EVALUATION OF THE MENTAL HEALTH PROFILE OF CAREGIVERS OF CHILDREN WITH CEREBRAL PALSY IN A LOW-RESOURCED SETTING: DEVELOPMENT, TRANSLATION AND VALIDATION OF PATIENT-REPORTED OUTCOME MEASURES

by

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a) **Dambi JM**, Corten L, Chiwaridzo M, Jack H, Jelsma J, Mlambo T. A systematic review of the psychometric properties of the cross-cultural translations and adaptations of the Multidimensional Perceived Social Support Scale (MSPSS). BMC Health and Quality of Life Outcomes; 2018; 16:1–19
b) **Dambi JM**, Jennifer J, Mlambo T, Chiwaridzo M. Use of patient-reported outcomes in low-resource settings — lessons from the development and validation of the Zimbabwean Caregiver Burden Scale. Eur J Physiother;2017: 19:sup1, 47-50
c) **Dambi JM**, Jelsma J, Mlambo T, Chiwaridzo M, Tadyanemhandu C, Chikwanha MT. A critical evaluation of the effectiveness of interventions for improving the well-being of caregivers of children with cerebral palsy: a systematic review protocol. BMC. Systematic Reviews; 2016; 5:112
d) **Dambi JM**, Jelsma J, Mlambo T, Chiwaridzo M, Dangarembizi-Munambah N, Corten L. An evaluation of psychometric properties of caregiver burden outcome measures used in caregivers of children with cerebral palsy: a systematic review protocol. BMC. Systematic Reviews; 2016; 5:42.
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Abstract

Background & aims: Several systematic reviews have demonstrated that caring for a child with functional limitations leads to poor caregiver mental health outcomes. For instance, depression, anxiety and caregiver burden are endemic in informal (unpaid) caregivers of children with cerebral palsy (CP), a common paediatric disability. However, caregivers who receive an adequate amount of social support are likely to better adjust to the caregiving role. Given the increasing move towards family-centred care and evidence-based care, there is a need to evaluate caregivers' mental health and to develop and implement context-specific caregiver well-being programs. The valid measurement of the impact of these programs is dependent on the availability of psychometrically robust patient-reported outcomes (PROMs). Unfortunately, most available PROMs have been developed in high-income settings, and their applicability in low-resource settings such as Zimbabwe may be questionable. The present study thus aimed to develop a context-specific caregiver strain outcome, to adapt, translate, and validate a social support outcome measure and finally to profile the mental health of caregivers of children with CP residing in Zimbabwe.

Methods: The Zimbabwean Caregiver Challenges Scale (ZCCS) was developed through the amalgamation of findings from a systematic review, caregivers' interviews and two rounds of content validation by a panel of experts. A systematic review was then undertaken to appraise the psychometric properties of the Multidimensional Scale of Perceived Social Support (MSPSS). The MSPSS was then adapted, translated into Shona, a Zimbabwean native language, and validated. In the final phase, further validation of the ZCCS and the MSPSS was done, and structural equation modelling was used to profile the mental health profile of caregivers.

Results: The ZCCS yielded four factors which were ; physical & economic burden, concerns for the child, family relations and community participation. The ZCCS was found to be a reliable tool as it yielded excellent Internal Consistency (IC) ratings at both sub-scale [α range: 0.765- 0.841] and scale level [α=0.925]. The Interclass Correlation (ICC) (95% CI) for ZCCS scores at baseline and after four weeks was 0.880 (0.793: 0.930), demonstrating the stability of the ZCCS. We replicated the original 3-factor structure of the MSPSS through factor analysis. Further, dichotomisation of scoring options and the deletion of one item resulted in a parsimonious solution as the 11-items solution met Rasch modelling requirements. The MSPSS-Shona version is reliable; it yielded excellent IC ratings at both sub-scale [α range: 0.833-0.892] and scale levels [α=0.8685]. The ICC rating (95% CI) for MSPSS scores at baseline and after four weeks was 0.980 (0.959: 0.990) demonstrating the stability of the MSPSS, and the person separation index (PSI) was 0.731. Both the ZCCS and MSPSS displayed construct

validity; higher caregiver burden was associated with greater psychiatric morbidity and lower health-related quality of life. Caregivers who received a higher amount of social support had the best mental health outcomes. Further, structural equation modelling provided evidence of the multidimensionality of the caregiver burden. Contextual factors, such as increased child age, increased caregiving duration, lower child functional capacity/more severe CP, and lower socioeconomic status were associated with poor mental health functioning.

Conclusion: The thesis contributes to the body of knowledge by documenting the validation of caregivers' mental health outcomes and determination of mental health functioning of caregivers residing in low-resource settings. The ZCSS and MSPSS were both found to be valid and reliable measures within the context of Shona speaking rural and urban Zimbabweans. It is therefore suggested that both measures can be used with confidence in these contexts. Efforts should be made to integrate patient-reported outcome measures (PROMs) in routine clinical care and research and in developing and implementation of tailor-made caregiver wellness programs, within the validation contexts.

Keywords: caregiver burden, social support, reliability, validity, Zimbabwe, cerebral palsy, mental health

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I dedicate this project to my daughter, Aviah Waishe Dambi, and my lovely wife, Nakai TN Dambi, for always being there for me through the ups and downs of the PhD journey.

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Abbreviations and glossary

ACEND	Assessment of Caregiver Experience with Neuromuscular Disease
AIC	Akaike's information criterion
ANOVA	Analysis of variance
APSP	Alberta perinatal stroke project parental outcome measure
ART	Anti-retroviral therapy
ASPI	Autism parenting stress index
BIC	Bayesian information criterion
CBI	Caregiver burden index
CBIn	Caregiver burden inventory
CBS	Caregiver burden scale
CCWCP	Caregivers of children with cerebral palsy
CDS	Caregiver difficulties scale
CFA	Confirmatory factor analysis
CFI	Comparative fit index
ChiSq	Chi-square
CMDs	Common mental disorders
Communality	Variance in observed variables which is accounted for by a common factor, it is the square of factor loading
Construct validity	The extent to which scores on a questionnaire relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are being measured
Content validity	The extent to which the domain of interest is comprehensively sampled by the items in the questionnaire
COSMIN	Consensus-based Standards for the selection of health status Measurement Instruments
CP	Cerebral palsy
Criterion validity	The extent to which scores on a questionnaire relate to a gold standard
CRU	Children rehabilitation unit
CSI	Caregiver strain index
CSQ	Caregiver strain questionnaire
CTT	Classical testing theory – is a measurement paradigm whereby a participant's score is the sum of their estimated ability and random error
CWDs	Children with developmental abilities
DF	Degrees of freedom

DIF	Differential item functioning
EBP	Evidence-based practice
EFA	Exploratory factor analysis (EFA)- is a statistical technique used to explore the number of factors an outcome measure possesses
Eigenvalue	Is the amount of variance explained by items within a factor
EPI	Expanded immunization program
EQ-5D	Euroqol five-dimension scale- generic health-related quality of life outcome measure
FAM	Family
FCDC	Fathers of Children with Developmental Challenges Questionnaire
FIBS	Family burden interview schedule
FICD	Family Impact of Childhood Disability
FIQ	Family impact questionnaire
FL	Factor loading - the correlation between a variable (item) and a factor
Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible score
FRE	Friend
GMFCS	Gross motor classification system - is a five-point ordinal measure for severity of CP with levels one and five being least and most severely affected individuals
GMFM	Gross motor function measurements
GOF	Goodness of fit
HCH	Harare central hospital
HIC	High-income country
HIV/AIDS	Human immunodeficiency virus/ acquired immune deficiency syndrome
HRQoL	Health-related quality of life
IC	Internal consistency
ICC	Item characteristics curves
ICF	International Classification of Functioning, Disability and Health
I-CVI	Item content validity index- the degree to which an item on an outcome measure measures the trait under investigation
Identity matrix	A matrix in which all off-diagonal elements are zero i.e. All correlation coefficients are equal to zero
Internal consistency (IC)	The extent to which items in a (sub)scale are intercorrelated, thus measuring the same construct
Interpretability	The degree to which one can assign qualitative meaning to quantitative scores

IPFAM	Impact on Family Scale
IQR	Interquartile range
IRT	Item response theory
ITC	Total-total correlation
KMO	Kaiser-Meyer-Olkin- a measure of the extent of sampling adequacy which determines whether data is suitable for factor analysis
Latent variable	A construct which cannot be directly measured but can be estimated by measuring an indicator/manifest/dependent variable
LCA	Latent class analysis
LFI	Tucker-Lewis index
LICs	Low-income countries
LMIC	Lower-middle-income country
LPA	Latent profile analysis
LTR	Likelihood ratio test
MBI	Maslach burnout inventory
M-CSI	Modified caregiver strain index
Mental Health	"… a state of well-being in which every individual realizes his or her own potential, can cope with the normal stresses of life, can work productively and fruitfully, and is able to make a contribution to her or his community" [17]. Mental health (MH) is a continuum of indices, in the present study, the term MH encompasses indices such as: caregiver burden, social support, depression, anxiety, health related-quantity of life and associated synonyms.
MIC	Middle-income country
MRCZ	Medical Research Council of Zimbabwe
MSPSS	Multidimensional Scale of Perceived Social Support- a 12-items social support outcome measures. Measures the amount of support friend from; family, friends and significant other/spouse.
NOSI	Nijmeegse ouderlijke stress index
Panel of experts	Refers to clinicians and academics with appropriate experience in the following domains; management of CP/paediatric disabilities, outcome measures development and mental health service provision and research. Experts were expected to exhibit at least two of the ascribed attributes/competencies.
Pattern matrix	Regression coefficients between variables and factors
PCA	Principal component analysis
PCDI	Parents of Children with Disabilities Inventory
PCM	Partial credit model
PedsQL	The Pediatric Quality of Life Inventory-Generic Core Scales
PI	Principal investigator

PRISMA	Preferred Reporting Items of Systematic Reviews and Meta-Analyses
PROM	Patient-reported outcome - patients' definition or conceptualisation of their health state [1][2].
PROSPERO	International prospective register of systematic reviews
PSI	Person separation index- an indicator of reliability under Rasch modelling, it is analogous to Cronbach Alpha
PSI	Parenting stress index
PSS	Parental stress scale
PSS	Perceived stress scale
QRS	Questionnaire on Resources and Stress
Rasch analysis	Is a probabilistic measurement model whereby a participants/ score (trait level) is a function of both item difficulty and respondent ability
RBM	Revised burden measure
RCT	Random controlled trial
Reliability	The degree to which the measurement is free from measurement error
Responsiveness	The ability of a questionnaire to detect clinically important changes over time
RMSEA	Root mean squared error of approximation
RSM	Rating scale model
S-CVI	Scale level content validity index
S-CVI/Ave	Scale content validity index-average
S-CVI/UA	Scale content validity index- universal agreement index
SD	The coefficient of determination/ standard deviation
SE	Standard error
SEM	Structural equation modelling
SES	Socio-economic status
SMCP	Stress Level of Mothers with Children with CP Measurement Tool
SO	Significant other
SR	Systematic review
SRMR	Standardised root mean squared residual
SS	Social support - the extent to which one receives support from the immediate family, society and healthcare professionals [3].
SSQ	Shona Symptoms Questionnaire – binary, 14-item mental health screening outcome measure
Structural/factorial validity	The extent to which scores on an outcome measure adequately reflect the dimensions/structure of the construct to be measured

Structure matrix	Correlation coefficients between variables and factors
TB	Tuberculosis
TBM	Teach-back method-interview technique whereby the researchers read out the questions and caregivers were requested to; synthesise the meaning of the construct and to repeat the question in their own words before responding
TDC	Typically developing children
Thematic analysis	Is a qualitative method for identifying, analysing, organising, describing, and reporting themes found within a dataset
ThrSE	Threshold standard error
UCT	University of Cape Town
UnCThr	Unstandardized threshold
USA	United States of America
WHO	World Health Organization
ZBI	Zarit burden inventory
ZCCS	Zimbabwean Caregivers Challenges Scale

1 Chapter 1: Introduction

1.1 Background and need

About 15% of the world population suffers from one form of disability with 80% of disabled people residing in low-income countries (LIMCs) [4]. Paediatric disability is particularly a primary global public health concern [5,6], and the estimated prevalence of paediatric disability is between 0.4-12.7 % in the middle- and LIMCs as compared to 2.8% in high-income countries [4]. Cerebral palsy (CP), the most common paediatric disability, has a high prevalence in low-resource settings [7,8]. Most children with CP present with functional limitations thus requiring life-long assistance from informal caregivers in performing activities of daily living [7,9-11]. Several systematic reviews have demonstrated that caring for a child with functional limitations leads to a gradual deterioration of caregivers' health [12-16]. Depression, anxiety, loneliness, low self-efficacy has also been reported in caregivers [17-23]. The global lifetime prevalence of common mental disorders (CMDs) is considerable in the general population (29.2%) and is ostensibly greater in caregivers of children with disabilities living in LIMCs [21,24-28]. Caring for a child with a life-long disability is envisaged as an "involuntary and circumstantial career" which is characterised by devoting substantial emotional, physical and economic resources to the care of the child by caregivers [7-10,29-32].

Given the crucial role played by informal (unpaid) caregivers in the upbringing and rehabilitation of a child with a disability (CWD), it is essential to evaluate and improve their mental health [13]. This is in keeping with the family-centred approach to care which emphasises the need to improve the mental health of caregivers of CWD in addition to the improvement of functional outcomes and HRQoL of CWD [33-36]. To this end, the World Health Organization (WHO) has put forward the Mental Health Action plan (2013-2020) [37,38]. One of the aims of the strategy is to "…strengthen information systems, evidence and research for mental health…" [39]. Improvement of caregiver mental health is dependent on the availability of psychometrically robust patient-reported outcome measures (PROMs) for screening and evaluative purposes [16,40,41]. A PROM is defined as the patients' definition or conceptualisation of their health state [1,2]. Measurement parameters can either be observable constructs, e.g. difficulty in walking, or latent/unobservable constructs, e.g. caregivers' perception of the challenges of caregiving [1]. The perception of caregiver challenges/burden is context-specific and dependent on socio-cultural influences [12,13,42,43]. This underscores the importance of the conceptualisation of caregiver challenges and the validation of mental health PROMs in caregivers residing in low-resource settings such as Zimbabwe.

1.2 Conceptual framework

It was necessary to have an in-depth understanding of the perceived impact of caring for a child with a developmental disability on caregivers' mental health and identify appropriate outcome measures. Consequently, the conceptual frameworks by Raina et al. [42] and the revised Wilson and Cleary HRQoL model by Ferrans et al. [44] were examined. Raina et al. postulated that caregiver burden/difficulties are an interaction between caregivers' background and context, child characteristics, intrapsychic factors, such as self-efficacy, and coping mechanisms/strategies, such as the amount of social support available [42,45]. Social support (SS) is defined as the extent to which one receives support from the immediate family, society and healthcare professionals [3]. It can range from emotional support and financial assistance to health education provision and supportive counselling services [46]. Caregivers with extensive social support networks are more likely to adjust better to the demands of caregiving [47]. Additionally, Wilson and Cleary conceptualised an individual's HRQoL as the interaction between an individual's characteristics, biological and physiological variables and the environment [44].

The study was thus carried out in three phases. In the first phase, the Zimbabwe Caregivers Challenges Scale (ZCCS), a context-specific and culturally acceptable tool to measure caregivers' perceived burden of care/caregiving challenges, was developed. Most caregiver mental health outcomes have been developed and validated in high-income countries (HIC), and their use in LICs may not be appropriate. The conceptualisation of latent variables such as caregiver challenges, social support and HRQoL is context-specific and is heavily dependent on socio-cultural background [25]. Issues such as cultural acceptability, feasibility and clinical utility need to be taken into consideration when developing context-specific outcome measures [25]. As there is a lack of standardised measures and common taxonomy in PROMs, [4] and no locally developed tool for screening caregiver stress was identified, it was necessary to develop a context-specific tool for measuring the challenges faced by caregivers.

Social support has been consistently demonstrated to be a critical buffer to caregiver burden [29,45,48-50]. However, no tool validated for use in the Zimbabwean context was identified. The Multidimensional Social Support Scale (MSSPS) was, therefore, adapted, translated into the local language, Shona, and validated. The MSPSS is one of the most extensively used and psychometrically sound tools in measuring social support [51-53]. The MSPSS has demonstrated trans-cultural validity and has been translated into several African native languages in Malawi [47], Uganda [3] and Nigeria [54]. Finally, the psychometric properties of the MSPSS-Shona version and ZCCS were validated, and

univariate analysis and structural equation modelling were done to determine the factors influencing caregivers' mental health within the Zimbabwean context.

1.3 Problem statement

It is known that caregivers of children with cerebral palsy are likely to report poorer mental health [13]. However, there is a paucity of large-scale studies done in the Zimbabwean context to evaluate caregivers' well-being. The lack of empirical evidence is worsened by a lack of psychometrically sound outcome measures which are context-specific and culturally sensitive.

1.4 Phase One: Development and preliminary psychometric evaluation of the ZCCS

1.4.1 Research questions, aims and objectives

Phase one set out to establish whether the newly developed ZCCS was a valid, reliable, and culturally acceptable tool for measuring caregiver challenges in Zimbabwean caregivers of children with CP. The specific objectives included the following:

- To develop the ZCCS, a tool which measures caregiver challenges based on a synthesis of literature, and from the views of; caregivers of children with CP and a panel of experts
- To determine the following psychometric properties of the newly developed ZCCS:
    - Face validity
    - Content validity
    - Cultural applicability
    - Structural validity
    - Internal consistency
    - Test-retest reliability

1.5 Phase Two: Adaption, translation and preliminary psychometric evaluation of the MSPSS

1.5.1 Research questions, aims and objectives

This phase was designed to answer the questions:

- Is the adapted and translated MSPSS tool culturally acceptable and psychometrically sound in measuring social support in caregivers of children with CP in the Zimbabwean context?
- Will items on the MPSSS measure a single latent trait (social support) as reported in the literature?

The aims were, therefore, to adapt, translate and validate the MPSS into Shona, a Zimbabwean native language and the specific objectives are listed below:

- To adapt and translate the MSPSS into the Shona language using the backwards-forward translation method
- To determine the following psychometric properties of the Shona version of the MSPSS;
    - Cultural applicability
    - Face validity
    - Structural validity
    - Internal consistency
    - Test-retest reliability
- To perform a Rasch analysis on the MSPSS to evaluate the following parameters:
    - Item location
    - Unidimensionality
    - Item invariance
    - Item and scale reliability

1.6 Phase Three: Further psychometric evaluation of study instruments and evaluation of caregivers' mental health

1.6.1 Research questions, aims and objectives

This phase aimed to evaluate further the psychometric properties of the ZCCS and the MSPSS, and by using these instruments, to determine the well-being of caregivers of children with CP. The research questions at this stage were:

- Can the ZCCS and MSPSS discriminate between two known groups, i.e. caregivers of children with minor health problems and caregivers of children with CP?
- What is the concurrent validity of the ZCCS when compared to the scores on the Caregiver Strain Index (CSI)?
- Is there a difference between caregivers of children with CP residing in Harare Metropolitan province and rural districts of Zimbabwe with regards to:
    - Perceived HRQoL, caregiver burden and social support levels?
    - Report on common mental disorders?
- What is the relationship between the following latent variables: caregiver challenges, social support, psychiatric morbidity and HRQoL?

- What are the covariates significantly associated with mental health functioning of caregivers of CWCP?
- What model would reasonably explain the mental health profile of caregivers of children with cerebral palsy (CWCP)?

The specific objectives were:

- To determine if there is a difference between caregivers of children with CP and caregivers of children with minor health problems with regards to;
    - Caregiver challenges/stress as measured by scores on the ZCCS and CSI
    - HRQoL as measured by scores on the EQ-5D
    - Social support as measured by scores on the MSPSS
    - The frequency of common mental disorders as measured by scores on the Shona Symptoms Questionnaire (SSQ)
- To evaluate the concurrent validity of the ZCCS by establishing if there is a correlation with scores on the CSI
- To evaluate the predictors of caregiver burden through univariate analysis
- To examine the relationship between caregiver burden, social support, psychiatric morbidity and HRQoL through structural equation modelling.

1.7 Justification and significance

To the author's knowledge, there is a dearth of large-scale, published literature on the impact of caregiving for a child with a physical disability in the Zimbabwean context. Findings from an earlier quasi-experimental study pointed to the increased caregiver stress and poorer HRQoL in caregivers [55]. However, study participants were drawn from an urban setting, and the well-being of caregivers of children with CP residing in rural areas was not assessed. This was a significant limitation as the rural population constitutes approximately 67% of the total population in Zimbabwe [56]. As most HRQoL research finds its origins in urban settings, rural area residents can be considered as a "neglected" population. This is rather unfortunate as a study on Sri Lankan caregivers of children with CP revealed that caregivers who resided in rural areas reported greater burden/stress when compared to those residing in urban areas [49].

Children with disabilities in Zimbabwe are among the most stigmatised and ill-treated groups [21], and it is likely that the brunt of stigmatisation is associated with poor mental health functioning in the caregivers. Previous studies have demonstrated the high incidence of depression in women in

Zimbabwe [57]. As most caregivers are women, it was essential to screen for psychiatric morbidity in this population to inform suitable interventions for mitigating the effects of long-term caregiving.

The socio-economic turmoil in Zimbabwe since 2000 left the health care system on the verge of total collapse [55]. However, the healthcare system is currently undergoing a gradual improvement and restructuring, and there is thus a need for research to guide policy in the structuring of services [28]. Findings from the present study could inform policy as it profiles caregivers' mental health which is important in the development of context-specific interventions. Further, with the competition for scarce funding for health care programs, the increased impetus towards integration of mental health programs, and the drive for evidence-based interventions, this study was expected to provide essential baseline data given that it is a large-scale survey profiling caregivers' mental health.

1.8 Outline of thesis

As shown in Table 1.1 (Page 28), the thesis is divided into three phases. Chapters one (Introduction) and two (Narrative literature review) provide background/contextual information, study research questions and objectives. Chapters three to six outlines the methodology and preliminary psychometric evaluation of the ZCCS. Chapter seven outlines the translation, adaptation and preliminary psychometric evaluation of the MPSSS. Chapter eight focuses on the further evaluation of the psychometric properties of the ZCCS and MSPSS scales and exploration of the determinants of caregiver well-being with Chapter nine concluding the study.

Table 1.1: Thesis outline

Background & contextual information		
Introduction	• Background information • Study objectives • Study setting	Chapter 1
Literature review	State-of-art narrative literature review	Chapter 2
Phase one: Development & preliminary psychometric evaluation of the ZCCS		
Systematic review	Identification & appraisal of the psychometrics of paediatric caregiver burden outcome measures	Chapter 3
Item generation	• Qualitative interviews with caregivers • A panel of experts' content validation • Cognitive debriefing	Chapters 4 & 5
Structural validation & reliability evaluation	• Field testing • Psychometric evaluation	Chapter 6
Phase two: MSPSS translation & preliminary psychometric evaluation		
Systematic review	Identification & appraisal of the psychometrics of MSPSS translations	Chapter 7
Translation	• Translation & adaptation of the MSPSS-Shona version • Cognitive debriefing	Chapter 7
Structural validation & reliability evaluation	• Field testing • Psychometric evaluation	Chapter 7
Phase three: Further validation of study instruments & determination of caregivers' mental health		
Further psychometric evaluation & mental health profiling	• Known-group, construct & concurrent validity testing • Univariate analysis • Structural equation modelling (SEM)	Chapter 8
Conclusion		
	• Synthesis of study outcomes • Study critique • Conclusions • Recommendations	Chapter 9

1.9 Study setting

1.9.1 Structure of Zimbabwean healthcare services

Study participants were drawn from Harare Metropolitan province and rural districts in Mashonaland Central and Mashonaland West Provinces. Zimbabwe has seven administrative provinces, of which six of them are predominantly Shona speaking. Every province has one tertiary, provincial hospital (which serves as the primary referral centre) and several secondary level district hospitals which are State owned. There are also missionary hospitals which are run mainly by churches. Regarding the referral chain, district hospitals are at the second level tier with community clinics, provincial hospitals and central hospitals constituting the primary, tertiary and quaternary levels respectively [28,56,58].

1.9.2 Rural areas research sites

Caregivers of typically developing children (TDC) in rural areas were recruited from primary care clinics and rural district hospitals' outpatient departments. Participants were also recruited from the community Expanded Immunization Program (EPI) sites. The EPI is a nation-wide program whereby healthcare practitioners travel and immunise children within their communities [58,59]. Caregivers of children with CP were recruited from the district hospitals' rehabilitation departments.

1.9.3 Urban areas research sites

In urban areas, caregivers of TDC were recruited from Harare City primary-care clinics. There are primary clinics in every residential suburb. Permission was granted to collect data from eight clinics; we conveniently selected clinics with the highest number of cases. Caregivers of children with CP were recruited from the Children Rehabilitation Unit (CRU) which is located at Harare Central Hospital (HCH). The CRU is the largest paediatric referral centre in Zimbabwe, and its catchment area(s) includes the entire Harare metropolitan province and national referrals [12,28]. We also recruited participants from the CRU peri-urban outreach sites [28].

2 Chapter 2: Narrative literature review

2.1 Aims and chapter outline

This chapter aims to give an overview of the impact of caring for a child with CP (a stereotypical paediatric disability) on caregivers' mental health. The first section outlines conceptual frameworks related to caregiver burden/challenges which are followed by a review of current evidence on caregiver burden and social support. The chapter concludes with an overview of the psychometric methods used for tool validation.

2.2 The International Classification of Functioning, Disability and Health as applied to Cerebral Palsy

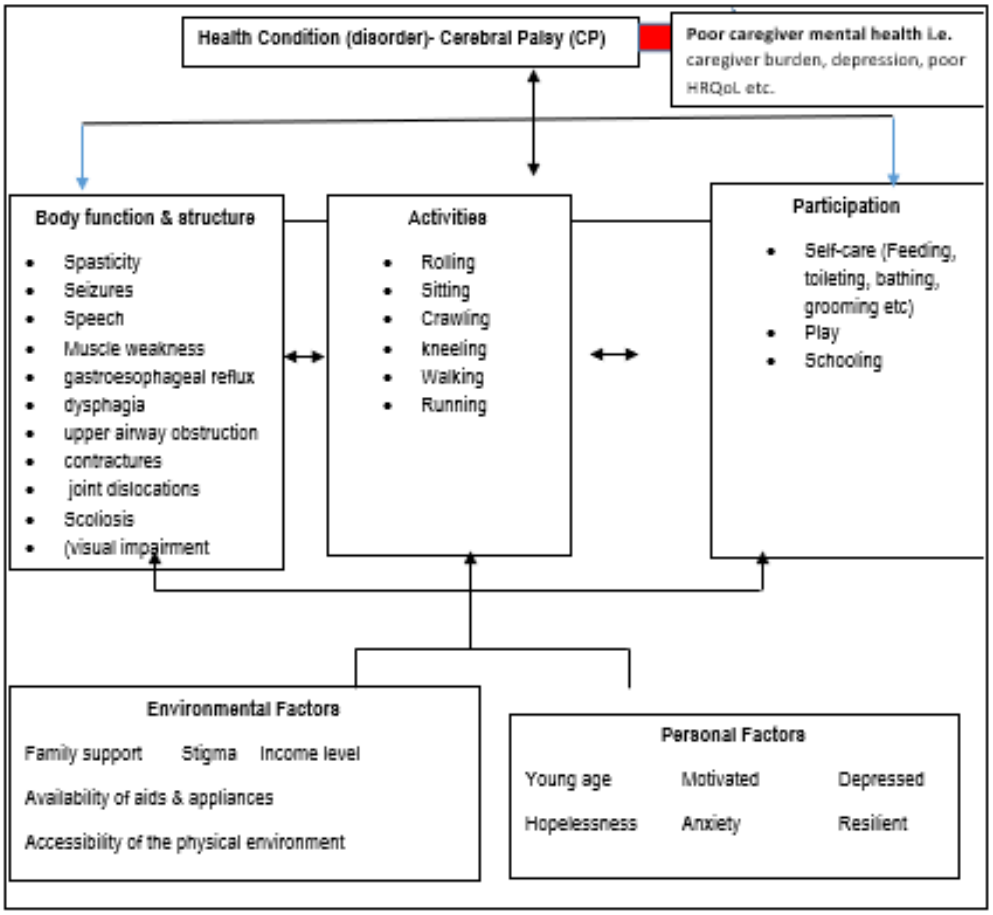
Cerebral palsy (CP) is a complex, stereotypical paediatric disability associated with functional limitations that require lifetime assistance in activities of daily living [9,20,60,61]. Unfortunately, caring for a child with CP very often leads to the deterioration of caregivers' physical and mental health which underscores the need to understand caregivers' lived experiences [62]. The World Health Organization's International Classification of Functioning, Disability and Health (ICF) is a useful model with which to conceptualise caregivers' experiences in caring for a child with a disability.

2.2.1 Background information on the ICF

The ICF is a multi-dimensional framework/classification system of health and functioning [63-75]. The ICF presents a paradigm shift in the conceptualisation of disability by departing from the bio-medical approach to disability [66-68]. Disability was previously envisaged as a linear process, i.e. alteration in physiological functioning led to a disability which was alleviated through medical interventions [63,64,68-70]. The ICF recognises the dynamic interaction between the health condition, impairments, activity limitations, participation restrictions, environmental factors and personal factors in tandem with the biopsychosocial model of disability [63-66,68-74]. In the context of paediatric rehabilitation, impairments imply any alterations at the anatomical or physiological level, e.g. seizures or increased tone [63,70,74]. Activity limitations imply a failure to perform functional activities such as rolling or sitting [63, 69-71]. Participation restrictions imply a failure to participate in daily life situations/activities such as play, schooling [63, 69-71]. Further, contextual factors, i.e. environmental and personal factors, also influence health and functioning [63,70]. Environmental factors include support structures (such as caregiver support), policies, societal attitudes and both natural and human-made changes to the environment [63,69,70]. Personal factors include variables such as age, coping factors, educational background and behavioural patterns, amongst others

[64,66,69.70,72,75]. Personal factors are further divided into positive (facilitators), e.g. motivation to learn and negative (barriers), e.g. hopelessness that can influence health states [69,70,71].

Using this framework, Figure 2-1 below depicts the possible multifactorial influences which may affect the overall functioning of a child with CP.



Sources; 66-90

Figure 2-1: The ICF framework as applied to a child with CP

2.2.2 Application of the ICF in the context of caregiving a child with CP

Cerebral palsy is a complex condition, with most children presenting with multiple impairments, activity limitations and participation restrictions [66,71,74,77,78]. CP can be clinically classified as ataxic, dyskinetic and spastic. The variants are hugely characterised by impairments in; balance and co-ordination, alteration in movement quality, and increase muscle tone respectively [66,71]. Nevertheless, motor impairments are the hallmark feature of CP, other impairments such as pain, seizures, visual disorders and mental retardation are common [60,79,80]. Impairments such as spasticity, muscle weakness, delay in movement initiation, poor force production and lack of muscle

coordination are likely to interfere with functional activities such as rolling, sitting, walking, among others [71,74,77,78,80]. Further, children with a higher number of impairments (a proxy indicator for severity of CP) are more likely to function poorly as predicted by the gross motor function classification system (GMFCS) [13,66,77,82-84]. Poor functioning will consequently result in reduced participation in daily activities such as play and schooling [66,71,74,77]. Environmental factors such as community attitudes may also influence the extent of disability and functioning [66,69,74,78,85]. For example, stigma by the community is likely to prejudice the child's ability to play/socialise with colleagues and this, in turn, will affect their motor functioning which consequently exacerbates impairments such as contractures which can arise because of physical inactivity thus perpetuating a vicious cycle [7,86-89]. More so, stigma is likely to affect the mental health of caregivers negatively [12,18,21,36,85,90]. Studies have shown that disabled children and their caregivers are amongst the most marginalised groups in low-resource settings [12,21,27,36,90-92]. Collective evidence from systematic reviews suggests that caregivers of CWCP are likely to suffer from caregiver burden, depression, anxiety, low self-efficacy and lower HRQoL [12,13,28,94-96]. The ICF framework clearly illustrates the multifactorial impact of childhood disability on the functioning of both the child and the caregiver. As the well-being of both child and caregiver are so inter-related, it is essential to monitor the impact of caring for a child with CP on the mental health of informal caregivers.

2.3 Caregiver burden/challenges

2.3.1 Introductions and definitions

Although there is general agreement that caring for a child with a disability is associated with caregiver strain/stress, there is a lack of consensus as to what constitutes the term "caregiver burden" [62,85,97-98]. The interchangeable usage of terms "caregiver stress", "caregiver strain", "caregiver challenges" and "caregiver distress" to express the experiences of caregivers is a clear testimony of the subjectivity and difficulty of conceptualising and operationalising caregivers' experiences [62,85,97,99,100]. Further, the use of the term "caregiver burden" may be associated with negative connotations and the expression "caregiver challenges" is envisaged as more acceptable terminology in depicting caregiver experiences in raising a child with a disability [101-103]. Nevertheless, most studies have used the term "caregiver burden" and, as such, the term will be used throughout this review to depict caregivers' challenges. Caregiver burden can be defined as the physical, psycho-social and financial effects of providing unpaid care to an individual with a long-term health condition [62,98,100]. Caregiver burden can be categorically classified as either an objective or a subjective burden [62,97,100,104]. Objective burden implies the "observable" effects of caring, e.g. increased

financial expenditure, whereas subjective burden reflects "unobservable" feelings associated with the caregiving experience, e.g. emotional outbursts [62,97,100,104].

2.3.2 Conceptual framework/theories

2.3.2.1 Introduction

Caregiver well-being is a complex and diverse phenomenon which has been subject to conflicting theories revolving around the subject area over decades [85,100,105,106]. This section reviews and synthesises some early and contemporary theories which explain caregiver burden and social support.

2.3.2.2 Double ABC-X Model

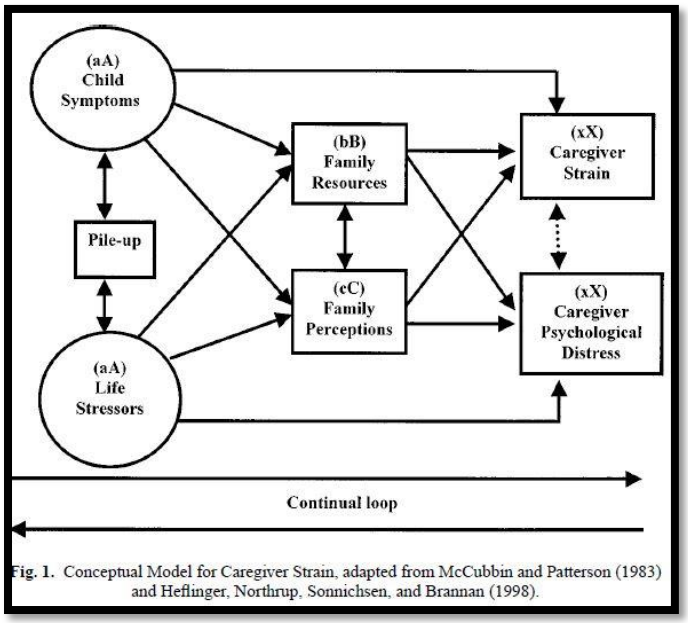


Fig. 1. Conceptual Model for Caregiver Strain, adapted from McCubbin and Patterson (1983) and Heflinger, Northrup, Sonnichsen, and Brannan (1998).

Source: [107]

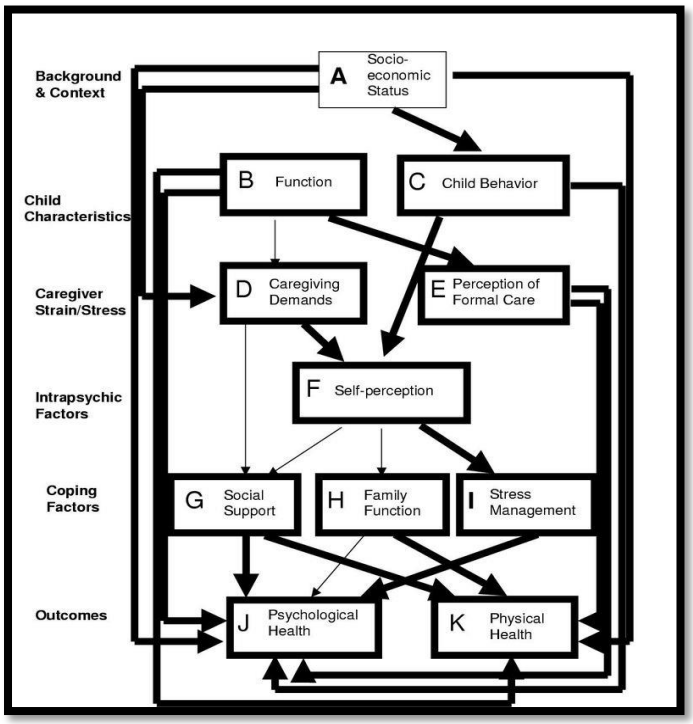
Figure 2-2: Double ABC-X Model

As illustrated in Figure 2-2 above, caregiver burden (xX) is a product of the continuous accumulation of stressors (aA), which negatively affect family resources/ability to deal with distress (bB) and family perception of the caregiving situation (cC) [107-109]. Stressors include both child characteristics, e.g. the presence of behavioural problems and stressful life events [99,107-109], which can be related to the ICF categories of impairments and functional limitations. Environmental factors, such as social support and the availability of professional support, are examples of family resources [99,100,107,108]. Family perceptions denote appraisal of the caregiving situation and attitudes towards available support structures [107,108]. The causal pathway is multidimensional with changes being cumulative as the perceived caregiver burden increases with the passage of time [99,100,107-109].

2.3.2.3 The transactional model of psychological stress and coping

Stress is viewed as a product of the environment and a person's appraisal of the "stressor"/stressful situation [105-107,110,111]. Stress occurs when individuals perceive that they are unable to handle the excessive demands of a stressor which threatens their well-being [105-107]. Cognitive appraisal and coping are significant predictors of a person's ability to handle a stressful situation [106,107,110, 112]. Cognitive appraisal is defined as the ability of a person to perceive any threat to their well-being and devise possible coping strategies to mitigate the stressor [105-107]. Coping refers to the cognitive (emotion-focused coping) and behavioural (problem-focused coping) efforts in dealing with the stressful situation [110-112]. Coping strategies can range from seeking social support, aggression, avoidance, self-control, escapism, denial, substance abuse and mental disengagement [105,111-113]. More stressful situations are often associated with the utilisation of diverse coping mechanisms, and this usually results in poor mental health and functioning [105,106,111-113].

2.3.2.4 Raina et al. conceptual framework



Source: [48]

Figure 2-3: Caregiver burden conceptual model

Raina et al. [48] postulated that caregiver burden is an interaction between five (5) distinct latent constructs, a model that is congruent with the ICF framework [63-75]. Caregivers' background/context influences the amount of strain; for example, low socioeconomic status is likely to be associated with a higher burden [48,98]. These are analogous to environmental and personal factors under the ICF

[63]. Secondly, child characteristics, particularly the severity of disability and the presence of behavioural problems, are cited as key predictors of caregiver burden [48,98] (impairments and functional limitations). Intrapersonal factors such as self-efficacy and mastery of the caregiving role also affect caregivers' perception of the caregiving role [98] (personal factors). Additionally, coping strategies such as social support, family function and pro-active stress management strategies are also likely to buffer the effects of caregiving [48] (environmental and personal factors). Therefore, caregiver burden is conceptualized as a dynamic interaction of caregivers' background and context, child characteristics, intrapsychic factors and coping mechanisms/strategies [42,48,98].

2.3.2.5 Synthesis of conceptual frameworks

Although there are divergent views with regards to the definition of the term "caregiver burden", there is a consensus that it is a subjective and multidimensional construct [62,85,97,99,100]. Caregiver burden emerges as a product of the child's characteristics, severity of disability, caregivers' internal resources such as self-efficacy, appraisal of caregiving situation and resilience, availability of both tangible and non-tangible social support resources and duration of the caregiving process [42,48,98-100,105-107,111-113].

2.3.3 Caregiver burden and paediatric disability narrative review

2.3.3.1 Search strategy and study selection

A state-of-the-art review [114] was performed by critically appraising literature published over the past five (5) years. The aims were to appraise the current evidence on caregiver burden and social support and outline determinants to caregiver well-being. This information was used in modelling caregivers' mental health profile (See Chapter Eight). The following databases were searched from January 2013 to 30th April 2018: PubMed, CINAHL, Scopus, PsycINFO and Africa-Wide Information. We utilised the following Boolean terms in searching for articles: ("caregiver" OR "care*" OR "mother") AND (("burden" OR "strain" OR "stress" OR "distress") OR ("social support" OR "social network" OR "social connections" OR "social relationships" OR "social isolation")) AND ("CP" OR "cerebral palsy" OR "disabilit*" OR "long-term health condition") AND ("child" OR "paediatr*").

The initial search yielded 72 articles, and we excluded articles which were not published in English, qualitative studies, commentaries, systematic reviews, and case studies. Data were extracted from 18 studies, and we utilised the Joanna Briggs Institute checklists [62] to rate the methodological quality/risk of bias of the studies. An overall synthesis is presented in Section 2.3.3.4 (Page 44) .

2.3.3.2 Methodological assessment of retrieved studies on caregiver burden

The methodological assessment of the studies is presented in the following three tables, stratified by the study design, i.e. cross-sectional (Table 2.1- Page 36), longitudinal (Table 2.2- Page 37) and quasi-experimental (Table 2.3- Page 38).

Table 2.1: Methodological assessment of retrieved studies-caregiver burden: Cross-sectional design

Authors (Year)	Country-Income bracket	Sample	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
Basaran et al. - 2013 [115]	Turkey – UMIC	1. Caregivers of children with CP, n=143 2. Caregivers of healthy children, n=60	-	-	+	?	-	-	+	+
Carona et al. - 2013 [116]	Portugal – HIC	1. Parents of children with CP, n=105: 2. Parents of healthy children, n=117	+	+	+	+	+	-	+	+
Carona et al. – 2013[117]	Portugal – HIC	1. Parents of children with CP, n=93: 2. Parents of healthy children, n=117	+	-	+	+	+	+/-	?	+
Carona et al. - 2014 [104]	Portugal – HIC	Parents of child with epilepsy (n = 65) and cerebral palsy (n = 91)	+	-	+	?	-	-	+	+
Wijesinghe et al. – 2015 [118]	Sri Lanka-LMIC	Caregivers of children with CP, N=375	+	-	+	?	-	-	+	+
Prihadi et al. - 2015 [113]	Netherlands-HIC	Parents of children with acquired brain injury, N= 42	+	-	?	?	+	-	?	-
Krstic´ et al. – 2015 [99]	Serbia-UMIC	Mothers of children with CP, N=100	+	-	+	?	+	+	?	+
Malm-Buatsi et al. -2015 [119]	USA-HIC	Caregivers of children with spina bifida, N=84	-	-	?	+	-	-	+	+
Prakash et al. - 2016 [120]	India-LMIC	Caregivers of children with CP, N=62	-	+	-	+	-	-	+	+
Lima et al. - 2016 [121]	Brazil – UMIC	Caregivers of children with CP, N=100	+	+	+	?	-	-	?	+
Chiluba & Moyo- 2017 [90]	Zambia-LMIC	Caregivers of children with CP, N=25	-	+	?	?	-	-	?	-
Kayadjanian et al. – 2018 [122]	USA-HIC	Caregivers of children with Prader-Willi syndrome (PWS), N=142	-	-	?	?	-	-	+	+
Karp et al. - 2018 [123]	USA-HIC	Parents of children with autism spectrum disorder (ASD), N=147	+	-	+	?	+	+	+	+

Key: Q1. Were the criteria for inclusion in the sample clearly defined?
Q2. Were the study subjects and the setting described in detail?
Q3. Was the exposure measured in a valid and reliable way?
Q4. Were objective, standard criteria used for measurement of the condition?
Q5. Were confounding factors identified?
Q6. Were strategies to deal with confounding factors stated?
Q7. Were the outcomes measured in a valid and reliable way?
Q8. Was an appropriate statistical analysis used?
From Joanna Briggs Institute checklists [62]

Table 2.2: Methodological assessment of retrieved studies-caregiver burden: Longitudinal design

Authors (Year)	Country-Income bracket	Sample	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11
Dambi et al. - 2014 [124]	Zimbabwe-LIC	Caregivers of children with CP, N=46	+	N/A	+	+	-	N/A	+	N/A	N/A	N/A	+
Difazio et al. - 2016 [125]	USA-HIC	Caregivers of children with CP, N=44	N/A	N/A	+	-	-	N/A	+	+	+	N/A	+
Vessey et al. – 2017 [126]	USA-HIC	Caregivers of children with CP, N=52	N/A	N/A	+	-	-	N/A	+	N/A	N/A	N/A	+

KEY: Q1. Were the two groups similar and recruited from the same population?
Q2. Were the exposures measured similarly to assign people to both exposed and unexposed groups?
Q3. Was the exposure measured in a valid and reliable way?
Q4. Were confounding factors identified?
Q5. Were strategies to deal with confounding factors stated?
Q6. Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?
Q7. Were the outcomes measured in a valid and reliable way?
Q8. Was the follow-up time reported and sufficient to be long enough for outcomes to occur?
Q9. Was follow up complete, and if -t, were the reasons for loss to follow up described and explored?
Q10.Were strategies to address incomplete follow up utilised?
Q11.Was an appropriate statistical analysis used?
From Joanna Briggs Institute checklists [62]
+=Yes, -=No, ?= Unclear

Table 2.3: Methodological assessment of retrieved studies-caregiver burden: Longitudinal design

Authors (Year)	Country-Income bracket	Sample	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9
Ferre et al. -2015 [127]	USA-HIC	Parents of children with CP, N=11	+	N/A	N/A	-	+	N/A	N/A	+	+
Raj et al - 2015 [128]	USA- HIC	Caregivers of children with mild traumatic brain injury, N=37	+	+	+	+	+	N/A	+	+	+

KEY:
Q1. Is it clear in the study what is the 'cause' and what is the 'effect' (i.e. there is - confusion about which variable comes first)?
Q2. Were the participants included in any comparisons similar?
Q3. Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?
Q4. Was there a control group?
Q5. Were there multiple measurements of the outcome both pre- and post the intervention/exposure?
Q6. Was follow up complete and if -t, were differences between groups in terms of their follow up adequately described and analysed? Q7. Were the outcomes of participants included in any comparisons measured in the same way?
Q8. Were outcomes measured in a reliable way?
Q9. Was an appropriate statistical analysis used?
From Joanna Briggs Institute checklists [62]
+=Yes, -=No, ?= Unclear

2.3.3.3 Outcomes of individual studies on caregiver burden

The results of the studies regarding impact are presented in Table 2.4 below.

Table 2.4: Results of retrieved studies-caregiver burden

Authors (Year)-Ref.	Country-Income bracket	Design	Sample	Child age (Years)	Outcome- standardized outcome measure	Proportion of caregivers reporting caregiver burden/ major (key) outcomes	Factors associated with elevated caregiver burden
Prakash et al. -2016 [120]	India-LMIC	Cross-sectional	Caregivers of children with CP, N=62	6.0 (SD 4.5)	1. Caregiver burden - Caregiver Strain Index (CSI) 2. Child functioning - Gross Motor Function Classification System (GMFCS)	77% of caregivers reported high levels of caregiver stress	More severe type of CP
Karp et al. -2018 [123]	USA-HIC	Cross-sectional	Parents of children with autism spectrum disorder (ASD), N=147	2-5	Parental stress - Parenting Daily Hassles survey	Caregivers reported high caregivers' stress	
Basaran et al. -2013 [115]	Turkey – UMIC	Cross-sectional	1. Caregivers of children with CP, n=143 2.Caregivers of healthy children, n=60	1. CP – 8.6 (SD 4.3) 2. Healthy-7.7 (SD 4.1)	1. Caregiver burden – Maslach Burnout Inventory (MBI) 2. QOL- WHOQOL-BREF 3. Depression- Beck Depression Inventory (BDI) 4. Anxiety – Beck Anxiety Inventory (BAI) 5. Child functioning - Gross Motor Function Classification System (GMFCS)	Caregivers of children with CP reported elevated; burden, depression, anxiety and lower QOL.	More severe type of CP
Kayadjanian et al. – 2018 [122]	USA-HIC	Cross-sectional	Caregivers of children with Prader-Willi syndrome (PWS), N=142	15.6 (SD 12.8)	Caregiver burden - Zarit Burden Interview (ZBI)	•56% experienced elevated caregiver burden ·Caregiver burden increased with the passage of time ·Caregiving affected caregivers' social relationships in 89.9% of participants	Lower social support

Authors (Year)- Ref.	Country- Income bracket	Design	Sample	Child age (Years)	Outcome- standardized outcome measure	Proportion of caregivers reporting caregiver burden/ major (key) outcomes	Factors associated with elevated caregiver burden
Wijesinghe et al. – 2015 [118]	Sri Lanka- LMIC	Cross-sectional	Caregivers of children with CP, N=375	1-12	Caregiver burden – Caregiver Difficulties Scale (CDS)	·Caregivers reported elevated caregiver strain ·Social support buffered caregiver burden	·Living in a rural area ·lower income ·male child ·lower spousal social support ·more severe type of CP
Chiluba & Moyo- 2017 [90]	Zambia - LMIC	Cross-sectional	Caregivers of children with CP, N=25		Caregiver burden - Modified caregiver strain index (MCSI)	64% reported caregiver strain	None
Prihadi et al. - 2015 [113]	Netherlands- HIC	Cross-sectional	Parents of children with acquired brain injury, N= 42	13.6 (SD 4.8)	1. Caregiver burden - Caregiver Strain Index (CSI) 2. Coping - Utrecht Coping List (UCL) 3. Family functioning - Family Assessment Device (FAD) 4. Life satisfaction - Life Satisfaction Questionnaire 9 (LiSat-9)	·50% experienced elevated caregiver burden ·43% reported a lower life satisfaction ·38% reported unhealthy family functioning ·Higher family dysfunction associated with higher levels of strain and a lower quality of life	None
Lima et al. 2016 [121]	Brazil – UMIC	Cross-sectional	Caregivers of children with CP, N=100	6 (SD 3)	1. Parenting stress- Parenting stress index-short form (PSI-SF) 2. Social support - Medical Outcomes Study Social Support Survey (MOSSS) 3. child functioning – gross motor classification system (GMFCS)	·42% of caregivers in clinical distress range ·81% received adequate social support	·Being single/ Divorced ·lower income ·more severe CP
Carona et al. - 2014 [104]	Portugal – HIC	Cross-sectional	Parents of child with epilepsy (n = 65) and cerebral palsy (n = 91).	12.5 (2.8)	1. Caregiver burden - The Revised Burden Measure 2. QOL- WHOQOL-BREF	Higher caregiver burden associated with lower HRQoL (r=-.43, p<.01)	

					3. Behavioural Disengagement- COPE Inventory 3. Child's QOL- KIDSCREEN-10.		
Authors (Year)- Ref.	**Country- Income bracket**	**Design**	**Sample**	**Child age (Years)**	**Outcome- standardized outcome measure**	**Proportion of caregivers reporting caregiver burden/ major (key) outcomes**	**Factors associated with elevated caregiver burden**
Carona et al. - 2013 [116]	Portugal – HIC	Cross-sectional	1. Parents of children with CP, n=105 2. Parents of healthy children, n=117	12.0 (SD 2.9)	1. Caregiver burden - The Revised Burden Measure 2. QOL- WHOQOL-BREF	·Caregivers of children with CP reported greater burden ·Higher caregiver burden associated with lower HRQoL	Increased child age
Krstic´ et al. – 2015 [99]	Serbia- UMIC	Cross-sectional	Mothers of children with CP, N=100	2-7	1. Family stress- Family Inventory of Life Events and Changes (FILE) 2. Depression- Depression scale (DS) 3. Child functioning - Functional status (FS II (R)) 4. Reaction to Diagnosis Interview (RDI)	·25% of mothers were depressed ·Correlations between parental stress and: i. Child functional status, r=-.36, p<.01 ii. Depression, r=.271, p<.01 iii. Lack of resolution of the child's diagnosis, r=-.219, p<.01	·Increased expenses ·lower educational level ·severity of CP
Carona et al. – 2013 [117]	Portugal – HIC	Cross-sectional	1. Parents of children with CP, n=93 2. Parents of healthy children, n=117	8-18	1. Caregiver burden - The revised burden measure. 2. Social support - Satisfaction with social support scale. 3. Psychological maladjustment - Mental Health Inventory – short form (MHI-5). 4. Child psychological adjustment - Strengths and Difficulties Questionnaire (SDQ). 5. Quality of life - (WHOQOL) – 8-item index (EUROHIS-QOL). Child's QOL- KIDSCREEN-10.	Relationship between caregiver burden and: i. Social support: r=-.35, p<.01 ii. QOL; r=-.36, p<.01 iii. Psychological maladjustment: r=.51, p<.01	-younger caregiver age

Authors (Year)- Ref.	Country- Income bracket	Design	Sample	Child age (Years)	Outcome- standardized outcome measure	Proportion of caregivers reporting caregiver burden/ major (key) outcomes	Factors associated with elevated caregiver burden
Malm-Buatsi et al. - 2015 [119]	USA- HIC	Cross-sectional	Caregivers of children with spina bifida, N=84	11 (SD 6)	1. Parental stress -PSI/SF 2.Depression-Beck Depression Index (BDI) 3.Anxiety- Beck Anxiety Inventory (BAI)	Correlation between caregivers' stress and: i. overprotectedness - r>0.6 ii. depression – r < 0.3	-younger caregiver age -not being involved in counselling -
Vessey et al. – 2017 [126]	USA- HIC	Longitudinal	Caregivers of children with CP, N=52	11.5 (SD 3.9)	1. Family stress - Impact on Family Scale 2. Experience with Neuromuscular Disease (ACEND) 3. Family expenses - The Family Expense Diary	76.9% of caregivers reported of financial burden	Increased caregiver age; lower income; Increased distance travelled to the hospital
Difazio et al.- 2016 [125]	USA- HIC	Longitudinal	Caregivers of children with CP, N=44	11.3 (SD 3. Vessey et al. – 2017 [123]7)	1. Caregiver stress - Assessment of Caregiver Experience with Neuromuscular Disease (ACEND) 2. Child's HRQoL - Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) 3. Child functioning - Gross Motor Function Classification System (GMFCS)	No changes in scores	N/A
Dambi et al. - 2014 [124]	Zimbabwe- LIC	Longitudinal	Caregivers of children with CP, N=46		1. Caregiver burden - Caregiver strain index (CSI) 2. HRQoL – EQ-5D-3L 3. Child functioning - Gross Motor Function Classification System (GMFCS)	50% experienced elevated caregiver burden Higher caregiver burden was associated with lower HRQoL; (Spearman's rho = −0.33, p = 0.027)	None
Ferre et al. - 2015 [127]	USA- HIC	Quasi-experimental; single-	Parents of children with CP, N=11	2.5- 4.5	1. Parenting stress- Parenting stress index-short form 2. Child's hand function - The Assisting Hand Assessment (AHA)	The intervention did not influence caregivers stress	n/a

		group design			3. Treatment goals - Canadian Occupational Performance Measure (COPM)		
Authors (Year)- Ref.	**Country- Income bracket**	**Design**	**Sample**	**Child age (Years)**	**Outcome- standardized outcome measure**	**Proportion of caregivers reporting caregiver burden/ major (key) outcomes**	**Factors associated with elevated caregiver burden**
Raj et al. --2015 [128]	USA- HIC	Random controlled trial	Caregivers of children with mild traumatic brain injury, N=37	3-9	1. Parenting stress- Parenting stress index- short form. 2. Parent mental health functioning- Global Severity Index (GSI) & Center for Epidemiological Studies Depression Scale (CES-D) 3. Parenting self-efficacy - Self-Efficacy Scale (CSES)	- no differences in parenting stress at baseline and end of the intervention	-Lower income -Lower education -Lower parental self-efficacy

2.3.3.4 Synthesis of the level of evidence on caregiver burden

Most of the studies were cross-sectional and originated in high-income settings. Caregivers of children with developmental disabilities reported higher levels of caregiver burden, depression and anxiety and lower HRQoL. The following factors were associated with increased caregiver burden; caring for a child with severe functional limitations, lower income, increased caregiver age, lower social support, living in rural areas, being single/divorced, lower education, lower self-efficacy, lower spousal support, increased caregiving period and increased child age [90,99,104,104,106, 113,115-128].

2.4 Social support and paediatric disability narrative review

2.4.1 Introductions and definitions

Social support (SS) has been postulated as an essential buffer to stressful life events such as caring for a child with a long-term condition [46,100,126,130]. SS optimises both psychosocial and physiological reactions to stressful life events [46,130]. For instance, having an adequate amount of SS has been demonstrated to be linked with biomarkers such as lower cortisol levels, lower blood pressure and high pain threshold in the face of adverse events [46]. Although not universally conceptualised and defined, SS can be defined as the amount of assistance one gets through interactions with other [131,132]. The support can be emotional (e.g. empathy), tangible (e.g. practical help) or informational (e.g. advice) [131-134].

2.4.2 Methodology for narrative literature review

We performed a state-of-the-art review [114] by critically appraising literature published over the past five (5) years [See section 2.3.3.1 for the search strategy]. The initial search yielded 208 articles, and we excluded articles which were not published in English, qualitative studies, commentaries, systematic reviews, and case studies. Data were extracted from 12 cross-sectional studies, and we utilised the Joanna Briggs Institute checklists [62] to rate the methodological quality of the studies (See Table 2.5, Page 45). Results for the individual studies are presented in Table 2.6 (Page 46), and an overall synthesis is presented in Section 2.4.5 (Page 49).

2.4.3 Methodological assessment of retrieved studies on social support

Table 2.5: Methodological assessment of retrieved studies-social support

Authors-Year	Country-Income bracket	Sample	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8
Al-Gamal & Long- 2013 [135]	Jordan – UMIC	Caregivers of children with CP, N=204	-	-	?	+	+	-	+	+
Jeong et al.- 2013 [136]	South Korea-HIC	Mothers of children with CP, N=181	-	-	?	+	+	+	-	+
Fiss et al. – 2014 [137]	Canada & USA – HICs	Caregivers of children with CP, N=398	+	+	+	+	+	-	+	+
McConnel et al. – 2014 [138]	Canada- HIC	Families of children with disabilities and behaviour problems, N=538	-	-	+	+	+	+	+	+
Pfeifer, L et al. - 2014 [139]	Brazil – UMIC	Caregivers of children with CP, N=50	-	?	+	+	-	-	+	+
Findler et al. – 2016 [140]	Israel – HIC	Caregivers of children with disabilities, N=191	-	?	+	+	+	+	+	+
Jayanath et al. - 2016 [141]	Malaysia – HIC	Caregivers of children with CP, N=398	+	+	-	+	+	-	-	+
Lima et al. - 2016 [142]	Brazil – UMIC	Caregivers of children with CP, N=	+	-	+	+	-	-	?	+
Ma & Mak – 2016 [143]	Hong Kong-HIC	Caregivers of children with physical disabilities, N=131	?	?	+	+	+	+	+	+
Frishman et al. - 2017 [144]	USA-HIC	Caregivers of children with a childhood-onset dystrophinopathy, N=191	-	+	+	+	+	+	+	+
Yu et al. – 2017 [145]	Taiwan- HIC	Caregivers of children with CP, N=63	+	-	+	+	-	-	+	+
Park et al. - 2018 [146]	South Korea-HIC	Caregivers of children with intellectual and developmental disability, N=172	-	-	-	+	+	+	-	+

KEY: Q1. Were the criteria for inclusion in the sample clearly defined?
Q2. Were the study subjects and the setting described in detail?
Q3. Was the exposure measured in a valid and reliable way?
Q4. Were objective, standard criteria used for measurement of the condition?
Q5. Were confounding factors identified?
Q6. Were strategies to deal with confounding factors stated?
Q7. Were the outcomes measured in a valid and reliable way?
Q8. Was an appropriate statistical analysis used**?**
+=Yes, -=No, ?= Unclear *(From Joanna Briggs Institute checklists [62])*

2.4.4 Results of retrieved studies on social support

Table 2.6: Results for retrieved studies- social support

Authors-Year [Ref.]	Sample	Child age (Years)	Outcome- standardised outcome measure	Major (key) outcomes	Factors associated with elevated social support
Al-Gamal & Long - 2013 [138]	Caregivers of children with CP, N=204	4.6 (SD not provided)	i. Social support (SS) - Multidimensional Scale of Perceived ii. Caregiver stress - Perceived Stress Scale (PSS10) iii. Beck Depression Inventory iv. Strengths and Difficulties Questionnaire v. Child functioning - Gross Motor Function Classification System (GMFCS)	i. Caregivers reported of good SS – mean MSPSS of 58.9 (SD 15.1) – the maximal score is 84 ii. Most SS was from significant other iii. Greater amount of SS associated with lower: child emotional and behavioral problems (r = 0.185, p = 0.04) iv. SS buffered effects of: parental stress (r = 0.389, p<.001) & depression (r = 0.379, p<.001)	None
Jeong et al. - 2013 [136]	Mothers of children with CP, N=181	<12	i. Social support - Carolina Parents Support Scale (CPSS) ii. Caregiver stress- Stress Level of Mothers with Children with CP Measurement Tool" (SMCP)	i) Social support mitigated the effects of caregiver stress, r=–0.260, p< .01 ii)Severe disability and lower education and income were associated with greater caregiver stress	Informal support mitigated stress more when compared to formal support received by mothers, (β= −1.117, p<0.01)
Fiss et al. – 2014 [137]	Caregivers of children with CP, N=398	3.7 (1.0)	i. Social support - Family Support Scale (FSS) ii. Family Support to Child (FSC) iii. Family Environment Scale (FES) iv. Family Expectations of Child (FEC) v. Child functioning - Gross Motor Function Classification System (GMFCS)	i. Social support was from multiple sources ii. The severity of disability did not predict the amount of SS received	None
McConnel et al. – 2014 [138]	Families of children with disabilities and behaviour problems, N=538	10.4 (SD 4.3)	i. SS- The health profile inventory ii. Family life congruence scale iii. Financial hardships scale iv. Developmental Behavior Checklist (DBC-24) v. General Family Functioning scale	i. Increased family congruency was associated with increased SS ii. SS buffered effects of caregiving	i. A mild form of disability ii. higher socioeconomic status

Authors- Year [Ref.]	Sample	Child age (Years)	Outcome- standardised outcome measure	Major (key) outcomes	Factors associated with elevated social support
Pfeifer, L et al. - 2014 [139]	Caregivers of children with CP, N=50	* 3-10 [Range]	i. SS- Social Support Questionnaire (SSQ) ii. Child functioning - Gross Motor Function Classification System (GMFCS)	i. SS was mainly from husbands and family ii. the severity of CP did not predict/affect SS received	None
Findler et al. – 2016 [140]	Caregivers of children with disabilities, N=191	3.3 (SD 1.3)	i. Social support (SS) - Multidimensional Scale of Perceived ii. Caregiver stress - Perceived Stress Scale (PSS10) iii. Subjective Happiness Scale iv. Handicap Related Events Checklist Symptom Severity Measure v. The Experiences in Close Relationships Scale (ECR)	• SS buffered the effects of i. stress- r=-.47, p<.001 ii. attachment avoidance- r=-.28, p<.05 iii. Attachment anxiety - r=-.47, p<.001 iv. guilt feelings • Greater SS associated with greater happiness	
Jayanath et al. - 2016 [141]	Caregivers of children with CP, N=398	10 (SD 5)	i. Social support (SS) - Multidimensional Scale of Perceived ii. Mental health - Depression, Anxiety and Stress Scale (DASS-21) iii. Child's HRQoL - Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD) iv. Child functioning - Gross Motor Function Classification System (GMFCS)	Caregivers reported of good SS – mean MSPSS of 63.2 (SD 13,1) – the maximal score is 84	None
Lima et al. - 2016 [142]	Caregivers of children with CP, N=398	6 (SD 3)	i. SS – Medical Outcomes Study Social Support Survey (MOS-SSS) ii. Stress- Parenting Stress Index (PSI) iii. Child functioning - Gross Motor Function Classification System (GMFCS)	i. 82% of caregivers were satisfied with SS received ii. SS did not buffer effects of stress	None
Ma & Mak – 2016 [143]	Caregivers of children with physical disabilities, N=131	12.9 (SD 4.0)	i. Social support (SS) - Multidimensional Scale of Perceived Social Support (MSPSS) ii. Caregiver worry - Parents of Children with Disability Inventory (PCDI)	Increased social support was associated with decreased: i. Worry; β=. -63, $p < .05$ ii. Stigma; β= -.66, $p < .05$ iii. Psychological distress; β= -.54, $p < .05$	Multiple sources of SS

			iii. Stigma - Affiliate Stigma Scale (ASS) iv. Psychological distress - Mental Health Inventory-18 v. Child functioning - Barthel Index (BI)		
Authors-Year [Ref.]	**Sample**	**Child age (Years)**	**Outcome- standardised outcome measure**	**Major (key) outcomes**	**Factors associated with elevated social support**
Frishman et al. -2017 [145]	Caregivers of children with a childhood-onset dystrophinopathy, N=191	16.1 (SD 6.5)	i. Social support (SS) - Multidimensional Scale of Perceived ii. Caregiver stress - Perceived Stress Scale (PSS10) iii. Family QOL - Beach Center Family Quality of Life Scale (FQoL) iv. Functional Assessment of Chronic Illness Therapy Spiritual Well-Being Scale (modified) (FACIT-Sp)	i. Caregivers reported of high SS ii. SS mitigated caregiver stress, ($r = -.29$, $p < .001$) iii. Increased SS was associated with increased QOL, ($r = .58$, $p < .001$)	i. increased family relationships ii. increase spirituality
Yu et al. -2017 [139]	Caregivers of children with CP, N=63	12 (SD 5.5)	i. Social Support Scale-modified (SSS-m) ii. Surgical concerns - Single-Event Multilevel Surgery (SEMS) scale iii. Child functioning - Gross Motor Function Classification System (GMFCS)	SS buffered caregivers' concerns for surgery ($r = - .434$, $p < .001$)	None
Park et al. -2018 [146]	Caregivers of youth with an intellectual and developmental disability, N=172	18.5 (SD 1.5)	i. Social support (SS) - Multidimensional Scale of Perceived Social Support (MSPSS) ii. Caregiver burden - Burden Assessment Scale (BAS) iii. Resilience - Connor-Davidson Resilience Scale (CD-RISC) iv. Coping - Brief COPE v. Stress - Perceived Stress Scale (PSS)	i) SS mitigated caregiver stress, ($r = -.52$, $p < .001$) ii) resilience & emotion-based coping mitigated caregiver stress iii) caregiver burden and the use of dysfunctional coping associated with greater caregiver stress	Less stigma

2.4.5 Summary of the review on social support

The present review confirmed SS as a buffer to adverse life events. An increased amount of SS was associated with decreased parental worry, psychological distress, stigma, attachment avoidance, depression, attachment anxiety, caregivers' concerns of treatment processes and guilt feelings. Further, increased SS was linked with increased HRQoL, family cohesion and happiness [135-146]. Multiple sources of SS, less stigma from society, coherent family relationships, increased spirituality, caring for a child with a mild disability, and being of high socio-economic status were cited as predictors of high SS [135-146].

2.4.6 Critique of the state-of-art narrative review

Findings from the present review need to be interpreted with caution as we did not reach literature saturation, and no backwards or forward searches were performed. Data were not independently extracted, and the risk of bias checklists used had several limitations. For example, the checklists did not evaluate the importance of sample size and ethical considerations in methodological quality. Additionally, methodological limitations such as lack of a clear selection criterion, use of outcomes with unknown psychometrics, inappropriate statistical analysis and a lack of adjustment for confounding may have negatively impacted studies' internal and external validity. However, the review was sufficient to answer the objectives of the thesis as it aimed to "appraise" the current evidence on caregiver burden and SS. Systematic reviews are recommended to understand the subject matter further.

2.5 Overview of psychometric evaluation methods

2.5.1 Introduction

As psychometric validation of both a new instrument (the Zimbabwean Caregiver Challenges Scale, ZCCS) and an existing instrument (the Multidimensional Scale of Perceived Social Support, MSPSS) were the primary aims of this thesis, it was essential to discuss how the validation of instruments may be performed. This section gives an overview of exploratory factor analysis (EFA), confirmatory factor analysis (CFA) and Rasch analysis psychometric evaluation methods.

2.5.2 Overview of classical testing theory (CTT)

Outlined in Table 2.7 (Page 50) are definitions of psychometric properties by the classical testing theory [147,148]. Under CTT, the psychometric robustness of an outcome is a combination of various validity and reliability indices [148-150]. Further, several indices can be used to give aggregated evidence of a specific psychometric property, for example; test-retest reliability, internal consistency,

and measurement error are all indicators of the reliability of an outcome [148-150]. These psychometric properties are prevalent in rehabilitation and validation literature; however, of these, structural validity requires sophisticated statistical computation and is also envisaged as the most critical psychometric property as it affects the quality of the rest of the psychometrical properties [151-157]. For instance, a tool can be reliable but not valid. However, a tool cannot be invalid and yield reliability [154,158]. We, therefore, explored different methods of structural validation in the subsequent sections, and these techniques were used in the validation of the ZCCS (See Chapter Six) and MSPSS (See Chapter Seven). More importantly, there is an increasing propensity towards the concurrent application of both CTT and item response theory (IRT) methods for structural validation [159-163], and this also necessitated an overview of both factorial validation methods.

Table 2.7: Definitions of psychometric properties

Property	Definition
1. Content validity	The extent to which the domain of interest is comprehensively sampled by the items in the questionnaire
2. Internal consistency	The extent to which items in a (sub)scale are intercorrelated, thus measuring the same construct
3. Criterion validity	The extent to which scores on a questionnaire relate to a gold standard
4. Construct validity	The extent to which scores on a questionnaire relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that
5. Reproducibility 5.1. Agreement	The extent to which the scores on repeated measures are close to each other (absolute measurement error)
5.2. Reliability	The extent to which patients can be distinguished from each other, despite measurement errors (relative measurement error)
6. Responsiveness	The ability of a questionnaire to detect clinically important changes over time
7. Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible scores
8.Interpretability	The degree to which one can assign qualitative meaning to quantitative scores

Source: Terwee et al. [147]

2.5.3 Exploratory Factor Analysis (EFA)

2.5.3.1 Introduction and definitions

EFA is a statistical technique used to explore the number of factors an outcome measure has [155-157,164-170]. Variables are grouped/lumped into factors which are defined as a cluster of highly correlated variables with a common underlying conceptual meaning [147-150]. Similarly, a factor can

be envisaged as a correlation matrix of highly correlated latent variables measuring the same construct [155,168]. Outlined below is some of the critical terminologies in EFA [155-157,159,164174]:

- Communality - variance in observed variables which is accounted for by a common factor, it is the square of factor loading
- Factor loading - the correlation between a variable (item) and a factor
- Identity matrix - a matrix in which all off-diagonal elements are zero, i.e. all correlation coefficients are equal to zero
- Pattern matrix - regression coefficients between variables and factors
- Structure matrix - correlation coefficients between variables and factors

2.5.3.2 Uses

EFA/common factor analysis can be used to reduce the number of items in a PROM while preserving content validity; this is referred to as parsimony [165-168]. Item reduction can be used to establish the dimensionality of a newly-developed PROM or translated PROM [165-169].

2.5.3.3 Requirements

To produce accurate results, data for EFA should be normally distributed, free of outliers and a valid factor should contain at least three (3) variables [165]. Further, the accuracy of the EFA solution is sample-size dependent [165]. Although there is a paucity of clear guidelines of "minimum sample size for EFA", a sample size of 300 is set as a bare minimum with some authors suggesting recruitment of 5-20 participants per item on a PROM [165,167,169].

2.5.3.4 Steps in performing EFA

Performing EFA is an iterative and subjective process [167]; however, an elemental analysis should include the following: testing of assumptions, factor extraction, factor rotation and interpretation [164-168].

- **EFA assumptions**

In addition to testing for normal data distribution and sample size, it is necessary to assess the extent of spread of respondents' responses, and this is termed sampling adequacy [171]. Adequate sampling adequacy is indicated by a Kaiser-Meyer Olkin Measure (KMO) value ≥ 0.5 [165,171]. The Bartlett's Test of Sphericity is also used to test the extent of the relationship between variables within a correlation matrix [167,171]. A significant relationship ($p< 0.05$) indicates suitability for data for EFA as patterns with variables will be significantly different from an identity matrix [165,171]. An identity

matrix is one in which the value of all the diagonal elements is one, such that if a correlation matrix is multiplied by the identity matrix, it remains unchanged. Consequently, the null hypothesis in EFA is that data is identical to the identity matrix [171].

- **Factor extraction**

Various methods can be used to extract factors, amongst these, the Kaiser's criterion of retention of all factors with an eigenvalue ≥ 1 is the most popular [165-168,171]. An eigenvalue is the amount of variance explained by items within a factor [166,171]. The Cattell scree plot which is a plot of eigenvalues against principal components can be used to extract factors [154]. Using the scree plot, all factors before the point of inflexion or break in the continuity of the plot are retained [165,168,171]. However, in some instances where the scree plot is difficult to interpret, alternative simulation methods such as Horn's parallel analysis method (PAM) can be used [155,166,169,171]. In PAM, random eigenvalues are generated, and only factors with random eigenvalues less than that of Kaiser eigenvalues are retained [155,166,169,171].

- **Factor rotation**

Factors are then rotated to improve interpretability as rotation aims to distribute the variance by maximising item loadings onto distinct factors [165,166,171]. Factors can undergo either oblique or orthogonal rotation [165,166,171]. The former is used when factors are hypothesised to be correlated which is the case of most PROMs [165,166,171].

- **Interpretation of factor loadings**

Given that factor analysis is based on the strength of linear correlations amongst variables, a factor loading of at least 0.30 is considered as a bare minimum [165,168,171]. Other authors advocate for 0.4 as the minimal as in correlation statistics, and r=0.4 is considered a moderate correlational strength [166,171]. A variable can load onto two or more factors; a phenomenon referred to as cross-loading [165,171]. Variables with cross-loadings of more than 0.32 on two or more factors are candidate items for deletion to improve interpretation of the factor solution [165,166,168]. However, it is essential to qualitatively analyse any item before deletion to preserve the content validity of a PROM [155,166,168,171].

- **Naming of factors**

The naming of putative factors is described as an "art" as there is a need to maintain simplicity and at the same time maintaining the conceptual meaning of items within the same factor [165,166,171].

2.5.3.5 Summary of EFA analysis plan

Outlined in Table 2.8 is the summary of the EFA plan as applied to this study [155,159,164,165,171-176].

Table 2.8: Exploratory factor analysis plan

Stage	Hypothesis/notes	Evaluative criteria
Testing of assumptions	Normality – data should be normally distributed	A non-significant Shapiro Wilkson Test statistic (p>0.05)
	Adequate inter-item correlations & absence of multicollinearity -	i. Adequate inter-item correlations, i.e. r≥0.3 ii. Items with excessively high inter-item correlations, i.e. r≥0.9 should be removed from the analysis iii. Item-total correlation; ITC≥.4
	Adequate sample variation	i. KM0≥ 0.5. [KMO values are interpreted as .5-.7- mediocre, .7-.8- good, .8-.9- great & >.9 – superb] ii. Participants to item ratio of at least 10:1
	Missing data	i. Missing values should occur in a non-random manner ii. Omit missing values to prevent over-estimation
	Factorability of the data	i. A significant Bartlett test of Sphericity- p≤ 0.05 means that the correlation matrices are statistically different from an identity matrix, i.e. there is a correlation between variables within the factor
Factor extraction Method	Various factor extraction methods can be applied, and they can yield different results	i. Try out several methods and report the parsimonious solution ii. Model selection is an iterative process and should be guided by theory
Dimensionality/ factor retention	Use multiple methods for factor retention	i. Kaiser's Criterion - retain factors with eigenvalues ≥ 1 ii. Horn's Parallel method - Create a set of random variables, and a factor is retained if the magnitude of the random eigenvalue is less that of Kaiser eigenvalue iii. Cartel's Scree plot- inspect for breaks in continuity on the eigenvalues vs. number of factors plot. Retain all factors just before the point of inflexion
Factor rotation	Apply oblique rotation	i. Compared to Promax, direct Oblimin rotation does not allow high correlation between factors ii. Apply both methods and select the parsimonious solution
Factor interpretation	Qualitatively analyse the conceptual meaning of items loading onto a factor	i. A meaningful factor should contain at least three (3) items ii. Factor naming is an "art."

2.5.4 Confirmatory factor analysis (CFA)

2.5.4.1 Introduction and definitions

CFA is an advanced structural equation modelling (SEM) statistical technique which combines the concepts of EFA, correlation, ANOVA and multiple regression [168,170,177,178]. It primarily involves testing the direction and strength of the relationship between multiple latent (unobservable) variables [171,178,179]. CFA is used to evaluate the degree of fit of a pre-existing factorial structure [170,171,178,179]. Modelling estimates are based on previous research or pre-set priori based on a sound theoretical framework where there is a direct contrast between the observed and estimated population covariance matrix [178,179].

2.5.4.2 Analysis procedure

Firstly, there is a need to specify the pathway diagram which provides the relationship between the observed (manifest/indicator) variables and unobserved (latent/common factor) [170,178]. A latent variable is defined as a construct which cannot be directly measured but can be estimated by measuring an indicator/manifest/dependent variable [170,171,178].

In the present case, items on the MSPSS would constitute observed variables with the three factors on the MSPSS denoting the unobserved variables. After that, the degree of model fit is tested against a pre-set criterion of fit indices (See Table 2.9 below) [170,178,180-186].

Table 2.9: Confirmatory factor analysis fit indices criterion

Fit statistic	**Index**	**Criterion for fit**
Likelihood Ratio	Chi-squared Test ($\chi ms2$)	p> 0.05
	$\chi 2/df$	<2
Population error	Root mean squared error of approximation (RMSEA)-(90% CI)	≤ 0.05
Information criteria	Akaike's information criterion (AIC)	Accept model with the lowest AIC value
	Bayesian information criterion (BIC)	Accept model with the lowest BIC value
Baseline comparison	Comparative fit index (CFI)	CFI≥0.90
	Tucker-Lewis index (LFI)	LT1≥0.90
Size of residuals	Standardised root mean squared residual (SRMR)	≤ 0.06
	The coefficient of determination (SD)	The greater the SD, the more useful the model

Multiple indices can be used to quantify the degree of model fit, and there is no consensus as to the combination of indices which can be used to assess a model [170]. The model can be modified thereafter should there be a model misfit. This is performed using modification indices [170,178]. However, caution needs to be exercised so that the modifications are based on sound theoretical justification as opposed to adjustment for the attainment of parsimony [168,170,177,178].

2.5.5 Rasch analysis

2.5.5.1 Introduction and definitions

Rasch analysis is a form of item response theory (IRT) structural validity and reliability analytical technique [187-196]. It has evolved as the "gold standard" in evaluating the structural validity of PROMs as it results in the conversion of ordinal scales to true metric (interval) scales [187-195]. Item difficulty/trait level (attribute being measured) is assessed on the same continuum with the persons' ability level, and both are measured in log-odds or logits [187-195]. For example, ability refers to the amount of perceived SS as measured by the MSPSS summative score whereas difficulty refers to participants' ranking of amount level of SS at the item level, i.e. a rating of 5 (strongly agree) on the 5-point Likert scale would be referred to as being difficult. Unlike classical testing theory (CTT) whereby a participant's score is the sum of their estimated ability and random error, in Rasch analysis measurement is envisaged as a function of both item difficulty and respondent ability [188,190-195].

2.5.5.2 Differences between IRT and CCT

There are fundamental differences between CCT and IRT [192-195,197, 206] and these are summarised below in Table 2.10.

Table 2.10: Differences between Item Response Theory & Classical Testing Theory

Attribute	**Classical Testing Theory**	**Item Response Theory**
Model	Linear	Non-linear
Assumptions	Few, i.e. easy to meet with test data	Difficulty in meeting model expectations
Test length	Lengthy scales yield the best validity and reliability estimates according to the Spearman-Brown Prophecy equation	Shorter scales can yield better validity and reliability indices
Response format	Items must have the same number of responses options (format)	Items may have an invariant number of responses options (format)
Local item dependency	A person's score on an item may depend on scores obtained from other items	Item scoring is independent across items

Item invariance	Items assumed to work the same for all study participants	Items can function differently between participants within the same sample (differential item functioning)
Sample invariance	Psychometrics may not be generalizable to other populations	Scale properties are sample independent
Person estimates	Scale-dependent	Scale independent
Item-person ability relationship	Not specified	Is a probabilistic relationship, information can be obtained from item characteristics curves
Data scale	Ordinal	Ordinal scales can be converted to true metric/ratio scale (additivity of scales)
Calculation of total sum score	The total score is a sum of the individual score and measurement error	The total score is the function of both item difficulty and person ability
Measurement error	Assumed to be constant across items	Varies across item difficulty
Reliability estimates	• Scores of persons with extremes responses/scores are included in the estimation of α • Usually high	• Extremes scores are extrapolated in the calculation of the PSI, a proxy of α • Usually lower than α
The sample size for item parameterisation	Usually in the 200-500 region	Depends on the model being used. Usually, 200 participants are adequate

Adapted from the following sources: [192-206]

2.5.5.3 Uses of Rasch analysis

Rasch analysis, a form of item response theory (IRT), is used to convert responses of PROMs from either nominal or ordinal level to true metric scale(s) [188-195]. True metric scaling is a prerequisite for the usage of parametric tests and for summing responses on PROMs graded on Likert-scaling [188-192,194,195]. IRT can thus be applied in developing a new PROM, reviewing/improving psychometrics of an existent PROM which would have been established through CTT, evaluating psychometric properties of a translated PROM, and in the development and calibration of item banks for computer adaptive testing (CAT) [188,190,193,195].

2.5.5.4 Steps in Rasch analysis

As with EFA, Rasch analysis is an iterative process; however, an excellent analysis should report on: model fit, scale targeting, threshold ordering, local dependency, dimensionality, differential item functioning (DIF), scale reliability and, where appropriate, scale repair [188-197] and these will be described in the proceeding paragraphs.

- **Model of analysis**

In Rasch analysis, the model is superior to the data, i.e. data is tested to evaluate if it meets model expectations [188,192,194,195]. Various models (dichotomous or polytomous) exist, and model selection is dependent on the nature of data and satisfaction of pre-requisite conditions [187,188,193,195]. One- and two-parameter logistic models are used for analysing dichotomous data whereas the partial credit model (PCM) and rating scale model (RSM) are used for polytomous data [187,189,191-193]. Polytomous models are utilised to analyse ordinal rated scales such as the MSPSS-Shona since it is scored on a five-point Likert scale. The decision to use either PCM or RSM is based on the outcome of a likelihood ratio (chi-square) test; if the test is statistically significant, the PCM is applied [188-190,193,195]. The RSM is used when the distances between categories are equidistant as evidenced by an ordered threshold. The PCM, on the contrary, does not have strict item stochastic ordering assumptions [188,192,193].

- **Category thresholds**

For polytomous items, there is a need to assess if there is a smooth and logical endorsement/discrimination of item category options by participants [188,189,192-194]. For a good model fit, it is expected that participants with higher trait levels will consistently endorse higher scoring options and vice-versa [188,192,193,195]. Additionally, for a well-ordered item, every category should have a point at which it has the highest probability to be endorsed by participants, inconsistencies in endorsements are referred to as disordered thresholds [187,189,192,195]. Disordered categories may be solved by iteratively combining adjustment categories until parsimony and model fit is attained [187,189,192,193,195].

- **Test of model fit**

As mentioned, in Rasch analysis, the data needs to be tested to establish if it fits the model. The model fit is assessed against a benchmark of various model fit parameters [187-189,192,195]. Various chi-square statistics are used to compare the observed and expected model fit [187,193,194]. An overall item-trait interaction statistic provides evidence of overall model fit; a significant chi-square statistic denotes model misfit; the null hypothesis is that data do not fit the Rasch model [188-193,195].

- **Test of item and person fit**

Both item and person fit statistics are transformed to standardised z-scores with an expected mean of zero (0) and standard deviation of one (1) [188,190,192,195]. Additionally, residuals in the range ± 2.5 denote both item and person fit [188,193,195]. Items can be further assessed for discrimination through usage of item characteristics curves (ICC), steep gradient(s) denotes over discrimination [188,190,192,193,195].

- **Item invariance**

In Rasch analysis, it is assumed that in the same population a scale should perform the same across different demographics, for example, male and female participants should ideally have the same probability of endorsing an item on a PROM [187-189,192,195]. Breach of this assumption is termed differential item functioning (DIF) [187-190,195]. The differences can be either systematic or non-systematic, and this is referred to as uniform and non-uniform DIF respectively [187-190,192,195]. Person factors such as age, gender and SES, amongst other pertinent demographic and clinical characteristics, can be used to assess for DIF [187-190,192,195].

- **Unidimensionality**

Unidimensionality is a prerequisite for Rasch scale modelling confirmation [187-190,195]. After extracting the Rasch factor through principal component analysis, a t-test is used to evaluate if there is a significant difference in residuals for the most positive and negative components [188,189,195]. To support unidimensionality, less than 5% of the tests should be statistically significant [189-192], or the lower boundary of an exact binomial confidence interval should be less than 5% [192,195].

- **Local dependence**

Local dependence is when two or more items are interlinked thus inflating classical reliability estimates and Rasch parameters [187,192, 195]. For example, a physical functional outcome assessing a patient's ability to walk with items asking the ability to walk distances of five (5) and 100 metres respectively. If a respondent can walk 100 metres, the respondent is likely to walk five metres without any difficulty and inclusion of both items would thus inflate the person's mean estimates [187,189]. Where there is a breach of local dependency, items can be either combined as a subtest or omitted to avoid redundancy [187,189,192,195].

- **Scale targeting**

Scale targetting is the extent of the spread of scores on a scale by assessing the mean item and person's scores [187,188,195,196]. A scale should not contain items that are either too difficult or too easy, and a scale is deemed well targeted if the mean person's location is near zero and 40-60% of the participant's scores are within three standard deviations of the zero-logit mark [187,189,192,195].

- **Scale repair**

Scale repair is undertaken if the preliminary analysis indicates model misfit. The repair process is an iterative process and often entails: rescoring of items to resolve disordered thresholds, the creation of sub-test lets/super-items to resolve the local dependency, stepwise deletion of misfitting items and removal of participants with extreme scores/responses [187-189,207].

2.5.5.5 Reliability

Scale reliability is assessed using the person separation index (PSI) which is analogous to the Cronbach's alpha [190,192,195,196]. However, the difference is that extreme scores are extrapolated in the calculation of the PSI whereas scores of persons with extremes responses/scores are included in the estimation of α [196]. A PSI ≥ 0.7 is deemed as evidence of good scale reliability [191-195].

2.5.5.6 Summary of Rasch analysis plan

Outlined in Table 2.11 below is the summary of the typical Rasch analysis plan [188-195]:

Table 2.11: Rasch analysis plan

Scale trait	Method of analysis	Hypothesis/Evaluative criteria
Partial credit (PC) vs rating scale (RS) parameters	Log-likelihood ratio	If RS parameters are satisfied (ratio test is non-significant), use RS format; if not, choose PC version
Response distribution	Frequency plot of actual responses across all items and response options	Check that each possible category of item/response is endorsed; ideal to have at least 5 cases in each
Thresholds	Graphic representation and plots of probability	Ordered progression of thresholds from less to more of the trait
Person fit	Chi-square, fit residual transformed to a standardised (Z) score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all items scored by a person
Item fit	Chi-square, fit residual transformed to a Z-score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all persons for a given item

Item trait interaction	Chi-square probability	p>0.05 tests whether the items are working as expected across the class intervals for the trait (the hierarchical ordering of items)
Individual person fit	Chi-square analysis	Values should fall within ±2.5 if the data fit the Rasch model
PSI	Cronbach's alpha	0-1; values over 0.70 allow for group comparison, over 0.85 for individual comparisons of summed scores
Local dependency	Correlation analysis of item residuals	Look for correlations of over 0.2 to 0.3
DIF	Item characteristic curves (IC curves); ANOVA of item residuals	IC curves plotted by person characteristics (such as age, sex, place of residence, SES, education) and p>0.05 for between groups ANOVA reveal DIF
Unidimensionality	Each subscale is partitioned using principal component factor analysis and subsequently t-tested	There will be no significant difference between the two partitioned pieces of the subscale

Adapted from the following sources: [188-195]

2.6 Conclusions of the narrative review

The narrative review introduced the ICF framework and, using this framework, explored the impact of a childhood disorder, specifically CP, on the child and the caregiver. It was concluded that the functional limitations associated with CP could be severe. The interaction between personal factors, functional limitations and environmental factors, which include caregiver support, necessitates a thorough understanding of the burden of caring for a child with a disability. The contested concept of "burden" was defined and analysed within different conceptual frameworks. The present review confirmed that caring for a child with a disability is associated with caregiver burden/caregiving challenges and caregivers who receive adequate SS are likely to cope with the caregiver strain. Child functional limitations and caregivers' self-efficacy, age, income level, education level, marital status, place of residence (urban vs rural), amount of family support, amount of spousal support and duration of caregiving influence caregivers' well-being.

The review concluded with an overview of the statistical techniques which can be used to establish the validity of PROMS, including a description of the steps required to undertake EFA, CFA and Rasch analysis. This section provides background to the systematic reviews of the psychometric properties of instruments designed to measure caregiver burden (See Chapter Three) and of the MSPSS (See Chapter Seven) presented in later chapters.

3 Chapter 3: A systematic review of the appraisal of psychometric properties of caregiver burden outcomes[1]

3.1 Introduction

Provision of care for a child with a long-term health condition is often associated with adverse health outcomes in caregivers, for instance, depression, stress, anxiety and low self-efficacy were reported in caregivers [12,13,28,94-96]. Cerebral palsy (CP) is the most common paediatric disability causing long-term functional limitations [7,8]. Children with CP most often present with multiple impairments, activity limitations and participation restrictions [9,79]. More so, due to its diverse and sophisticated presentation, CP is envisaged as the prototype paediatric disability [7,8]. As such, most children require extensive lifetime assistance in functional day-to-day activities [60,79,208]. The level of necessary assistance depends on the severity of impairments, activity limitations and participation restrictions [209]. Although taking care of a child is part of normal parenthood, the excessive demands associated with taking care of a child with a disability/long-term health condition may lead to increased burden/strain [12,25]. Consequently, long-term caregiving for a child with a disability such as CP may negatively affect the well-being of caregivers [96,2018,210].

Caregiver burden has been defined as "*strain or load borne by a person who cares for a family member with a disability"* [29]. Caregiver burden is multifactorial, complex, subjective and dynamic as envisaged in different conceptual models which have been developed to explain this construct [29,42,211,212]. The conceptual model by Raina et al. (2004) is one of the most cited and applied caregiver burden conceptual framework [42]. It postulates that caregiver burden is an interaction between the caregivers' background, contextual factors, child characteristics, intrapsychic factors and coping factors [42]. For instance, the presence of behavioural problems in children with CP, caregivers' socio-economic status, availability of social support and caregivers' self-efficacy all affect the overall perception of the burden of care [42]. Although usage of different semantics in describing caregiver burden makes it difficult to come up with a universally conceptualised definition and model, it is clear that long-term caregiving may lead to physical, psychological, emotional, social and financial strain [29,211,212].

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[1] The protocol for this study has been published. See Appendix 24
Dambi J, Jelsma J, Mlambo T, Chiwaridzo, Dangarembizi-Munambah N, Corten L (2016): An evaluation of psychometric properties of caregiver burden outcome measures used in caregivers of children with cerebral palsy: a systematic review protocol. Systematic Reviews 5:42: DOI 10.1186/s13643-016-0219-3
.

With the advent of patient-centred and family-centred approaches to clinical care, the need to evaluate services from patients' perspective, specifically the perceived impact of care on patients' well-being, becomes more important [213]. More so, it is essential to evaluate both physical and mental health outcomes in caregivers as they are an invaluable resource in the rehabilitation/treatment of children with long-term disabilities/health conditions [12,28]. For instance, the caregiver acts as the provider, decision maker, companion, custodian and advocator for a child with a long-term health condition [214]. Thus, routine assessment/screening of caregiver burden is of paramount importance for optimal functional outcomes of children with long-term health conditions.

Given the well-documented effects of caregiving on the health of caregivers, it is essential to routinely screen for caregiver burden/strain [12]. This is only attainable using psychometrically sound outcome measures [25]. According to the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) guidelines, an outcome measure must be valid, reliable and responsive for it to adequately capture the construct it is purposed to measure [149]. Over the past few decades, there has been an exponential increase in the number of outcome measures evaluating caregiver burden [25]. However, some of them are generic, and their utility in measuring the burden of care in caregivers of children with disabilities may be questionable [25,215,216]. Further, there is a paucity of systematic evidence of the psychometric robustness of tools measuring caregivers' burden while caring for a child with a disability.

Therefore, the broad objective of this systematic review was to evaluate the psychometrical properties of tools that have been utilised to measure caregiver burden in caregivers of children with CP. The specific objectives were to:

- Identify tools used to measure perceived caregiver burden in caregivers of children with CP
- Evaluate the psychometric properties of the identified outcome measurement tools
- To evaluate the clinical utility of the identified outcome measurement tools

3.2 Methods

3.2.1 Protocol registration

We used the Preferred Reporting Items of Systematic Reviews and Meta-Analyses (PRISMA) guidelines [217]. The protocol was registered on PROSPERO database (Ref: CRD42015028026) and has been previously published [218].

3.2.2 Eligibility criteria

In selecting the studies, the following criteria were applied:

3.2.2.1 Study designs/interventions

Precedence was given to articles on the development and validation of tools for measuring caregiver burden in caregivers of children with CP and/or other physical conditions such as spina bifida, hydrocephalus among others. Although CP is a prototype paediatric disability [7,8], it was hypothesized that the burden of caring might be equivalent, regardless of the causative condition. Therefore, other long-term conditions such as Down Syndrome, developmental delays, autism spectrum disorders, cancer among others were included. Studies that evaluated caregiver burden/strain in informal caregivers of children with long-term disabilities or interventional studies with caregiver burden as an outcome measurement were also included. Systematic reviews, qualitative studies, mixed study designs, case studies and editorial letters were evaluated. All quantitative study designs were considered to capture as much information as possible.

3.2.2.2 Participants

The changes in the dynamics of caregiving along the developmental trajectory were recognised. For instance, the dynamics of caregiving for a teenager with a disability may be different from providing care for a child in the age range 0-12 years [117]. Therefore, studies were included which examined the perceived burden of care in informal caregivers (18 years or older) of children between 0-12-year-old with long-term disabilities. An informal caregiver was defined as someone who takes the primary responsibility for giving care to a child with a disability and is not formally educated to take on nor remunerated for assuming the caregiver role.

3.2.3 Language

Full articles published in the English language only were considered as we did not have the financial resources to translate articles written in any other languages.

3.2.4 Information sources

The following databases were searched from their inception up to March 2018: PubMed, CINAHL, Scopus, PsycINFO and Africa-Wide information. To ensure literature saturation, where only abstracts were available, we contacted the authors to request the full text. If, after consulting the authors, only an abstract was available, the article was excluded. Grey literature was reviewed, and the Google Scholar search engine was used to search potential databases such as university databases,

conference proceedings for articles. For completeness, the reference lists of identified articles were also manually searched.

3.2.5 Search strategy

Outlined in Table 3.1 below is an example of how we searched for the articles in CINAHL database. As an illustration, we entered the following keywords to search articles in the CINAHL database: ("Caregiver" OR "care*" OR "mother") AND ("burden" OR "strain" OR "stress" OR "distress") AND ("outcome" OR "tool" OR "scale") AND ("valid*" OR "reliability" AND "dev*") AND ("CP" OR "cerebral palsy" OR "disabilit*" OR "long-term health condition") AND ("child" OR "paediatr*" AND " development" OR "construction").

Table 3.1: Search strategy

Keyword	**Alternative words**
Caregiver	Carer* OR mother OR parent* OR legal guardian*
Children	Child* OR paediatric* OR toddler* OR infants* OR pediatric*
Cerebral palsy	CP OR physical disability OR disability* OR neurodev* disorder*OR traumatic brain injur*
Burden	Strain OR stress OR burnout
Outcome measure	Tool OR questionnaire OR scale OR assessment
Psychometric	Validity OR reliability OR responsiveness
Evaluation	Determination OR measurement
Development	Construction

3.2.6 Data management

The articles were imported into RevMan (version 5.3) data management software. The electronic searches were also saved on users' PubMed, Scopus and EBSCOhost accounts. The summaries of all the searches were printed to enhance the data capturing of the search records. The principal investigator created a DropBox which acted as a repository for all the articles, and this was made available to all co-authors.

3.2.7 Data collection process

The principal author (JD) searched the databases and extracted titles and abstracts for further investigation. Thereafter, two researchers (CN and MC) independently retrieved the full manuscripts of articles considered relevant. Two other independent reviewers (LC & TM) blindly screened the

retrieved articles using a standardised data collection form. Information extracted included the research setting and design, study sample, demographics of the participants, mode of administration, number of items, cost, total score and the year in which the tool was developed. In case of a disagreement, a third reviewer (JJ) made the final decision.

3.3 Outcomes and prioritisation

For this review caregiver burden/strain, was the primary outcome measure. Secondary outcome measures included psychosocial indices such as depression, anxiety, stress, and self-efficacy.

3.4 Risk of bias in individual studies

The four-point COSMIN checklist was used to assess the methodological quality of the reviewed studies [149,150]. This was essential to prevent the risk of selecting and evaluating tools which were developed using designs with poor methodological rigour [149]. The COSMIN checklist rates the rigour of the reliability, validity, responsiveness, hypothesis testing, interpretability and generalizability of studies on the development and use of health-related patient-reported outcomes [149,150]. Items are rated on a four-point Likert scale, i.e. “excellent”, “good”, “fair” and “poor”. Where details are not published, the authors of the article were contacted to achieve the most truthful rating of the assessment tool and to decrease bias in the analysis.

3.5 Psychometric properties and data extraction

The psychometrical properties were evaluated using the checklist as outlined by Terwee et al. [147] (See Table 3.2- Page 66). Each psychometric property can be rated as positive, negative or questionable. An ideal tool should possess positive ratings [147].

Table 3.2: A quality criterion for psychometrical properties

Property	Definition	Quality criteria [a, b]
1. Content validity	The extent to which the domain of interest is comprehensively sampled by the items in the questionnaire	**+** A clear description is provided of the measurement aim, the target population, the concepts that are being measured, and the item selection AND target population and (investigators OR experts) were involved in item selection; **?** A clear description of above-mentioned aspects is lacking OR only target population involved OR doubtful design or method; **—** No target population involvement; **0** No information found on target population involvement.
2. Internal consistency	The extent to which items in a (sub)scale are intercorrelated, thus measuring the same construct	**+** Factor analyses performed on adequate sample size (7 * # items and >100) AND Cronbach's alpha(s) calculated per dimension AND Cronbach's alpha(s) between 0.70 and 0.95; **?** No factor analysis OR doubtful design or method; **—** Cronbach's alpha(s) <0.70 or >0.95, despite adequate design and method; **0** No information found on internal consistency.
3. Criterion validity	The extent to which scores on a questionnaire relate to a gold standard	**+** Convincing arguments that gold standard is ''gold'' AND correlation with gold standard >0.70; **?** No convincing arguments that gold standard is ''gold'' OR doubtful design or method; **—** Correlation with gold standard < 0.70, despite adequate design and method; **0** No information found on criterion validity.
4. Construct validity	The extent to which scores on a questionnaire relate to other measures in a manner that is consistent with theoretically derived hypotheses	**+** Specific hypotheses were formulated, AND at least 75% of the results are in accordance with these hypotheses **?** Doubtful design or method (e.g., no hypotheses); **—** Less than 75% of hypotheses were confirmed, despite adequate design and methods; **0** No information found on construct validity.
5. Reproducibility 5.1. Agreement	The extent to which the scores on repeated measures are close to each other (absolute measurement error)	**+** MIC < SDC OR MIC outside the LOA OR convincing arguments that agreement is acceptable **?** Doubtful design or method OR (MIC not defined AND no convincing arguments that agreement is acceptable); **—**MIC > SDC OR MIC equals or inside LOA, despite adequate design and method; **0** No information found on agreement.

5.2. Reliability	The extent to which patients can be distinguished from each other, despite measurement errors (relative measurement error)	**+** ICC or weighted Kappa > 0.70; **?** Doubtful design or method (e.g., time interval not mentioned); **—**ICC or weighted Kappa < 0.70, despite adequate design and method; **0** No information found on reliability.
6. Responsiveness	The ability of a questionnaire to detect clinically important changes over time	+ SDC or SDC < MIC OR MIC outside the LOA OR RR > 1.96 OR AUC > 0.70; **?** Doubtful design or method; **—**SDC or SDC > MIC OR MIC equals or inside LOA OR RR < 1.96 OR AUC < 0.70, despite adequate design and methods; **0** No information found on responsiveness.
7. Floor and ceiling effects	The number of respondents who achieved the lowest or highest possible score	**+** <15% of the respondents achieved the highest or lowest possible scores **?** Doubtful design or method; **—**> 15% of the respondents achieved the highest or lowest possible scores, despite adequate design and methods **0** No information found on interpretation.
8.Interpretability	The degree to which one can assign qualitative meaning to quantitative scores	**+** Mean and SD scores presented of at least four relevant subgroups of patients and MIC defined; **?** Doubtful design or method OR less than four subgroups OR no MIC defined; **0** No information found on interpretation.

Source: Terwee et al. [147]

3.6 Best evidence synthesis

Where a tool had been validated in several studies, the findings were combined to produce the best evidence for that tool. For this purpose, the previously established criterion for synthesising evidence by the Cochrane Collaboration Back Review Group [219] was used (See Table 3.3 below).

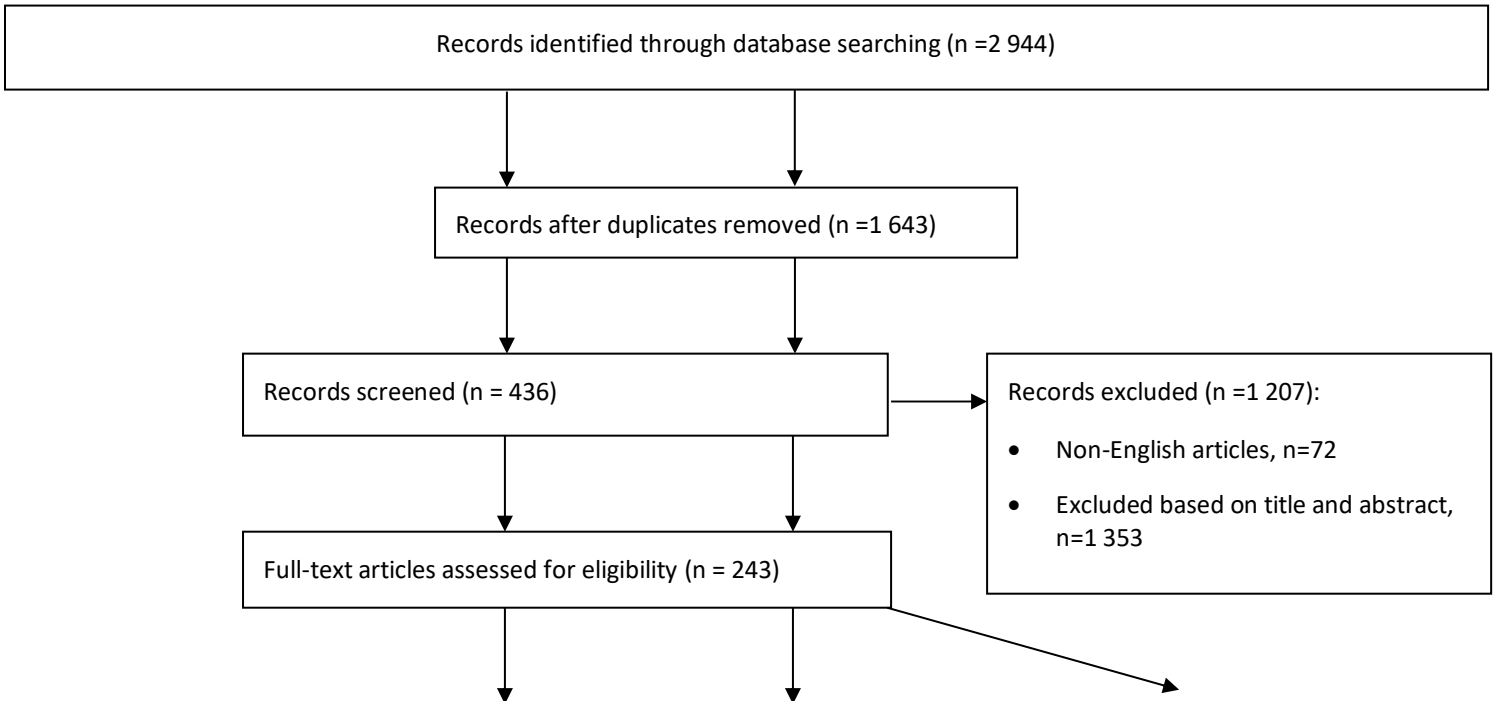
Table 3.3: Levels of evidence for the overall quality of the measurement property

Level	**Rating** [a]	**Criteria**
Strong	+++ or - - -	Consistent findings in multiple studies of good methodological quality OR in one study of excellent methodological quality
Moderate	++ or --	Consistent findings in multiple studies of fair methodological quality OR in one study of good methodological quality
Limited	+ or -	One study of fair methodological quality
Conflicting	+/-	Conflicting findings
Unknown	?	Only studies of poor methodological quality

[a]*Note that the evidence could point to either the acceptance or rejection of the instrument.*

3.7 Results

3.7.1 Study selection



No. of articles included in review (N=243) by tool:

1. Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM, n=3
2. Assessment of Caregiver Experience with Neuromuscular Disease (ACEND), n=3
3. Autism Parenting Stress Index (APSI), n=3
4. Caregiver Burden Index (CBI), n=2
5. Caregiver Burden Inventory (CBIn), n=1
6. Caregiver Burden Scale (CBS), n=4
7. Caregiver Difficulties Scale (CDS), n=3
8. Caregiver Strain Index (CSI), n=7
9. Caregiver Strain Questionnaire (CSQ), n=2
10. Family Burden Interview Schedule (FIBS), n=2
11. Family Impact of Childhood Disability (FICD)- original version, n=6
12. Family Impact of Childhood Disability (FICD)- revised version, n=1
13. Family Impact Questionnaire (FIQ), n=7
14. Fathers of Children with Developmental Challenges Questionnaire (FCDC), n=1
15. Impact on Family Scale (IPFAM)- original version, n=16
16. Impact on Family Scale (IPFAM)- simplified version, n=2
17. Maslach Burnout Inventory (MBI), n=4
18. Modified Caregiver Strain Index (M-CSI), n=3
19. Nijmeegse Ouderlijke Stress Index (NOSI), n=1
20. Parental Stress Scale (PSS), n=3
21. Parents of Children with Disabilities Inventory (PCDI), n=2
22. Perceived Stress Scale (PSS-4), n=4
23. Perceived Stress Scale (PSS-10), n=7
24. Perceived Stress Scale (PSS-14), n=12
25. PSI- Long Form (PSI-LF), n=9
26. PSI-Short Form (PSI-SF), n=87
27. Questionnaire on Resources and Stress- Short Form (QRS-SF), n=29
28. Revised Burden Measure (RBM), n=7
29. Stress Level of Mothers with Children with CP Measurement Tool (SMCP), n=1
30. The Pediatric Quality of Life Inventory-Generic Core Scales (PedsQL)- Family Impact Module, n=8
31. Zarit Burden Inventory (ZBI), n= 6

Figure 3-1 : Flowchart of article search and selection process

As demonstrated in the flow chart (Figure 3-1-Page 68), 2 944 articles were identified, of which 1 643 were duplicates. After applying the selection criterion, 243 articles were analysed for the present review. Most of the outcomes [29/31] were developed in high-income settings (Table 3.3).

3.7.2 Description of study participants and settings

Most of the outcomes [29/31] were developed in high-income settings. Depression, anxiety, HRQoL, and self-efficacy were the most common outcomes caregiver burden were contrasted with in evaluating construct validity (See Appendix 11.1- Page 236).

3.7.3 Description of outcome measure characteristics

Articles related to 31 outcome measures were retrieved and of these 23 were generic and most measures had 10-26 items [23/31], were rated on a 5-point Likert scale [21/31], were available for free [28/31] and had scoring instructions available [19/31]. The Parental Stress Index (PSI) & Questionnaire on Resources and Stress - Short Form (QRS-SF) were the most frequently used outcome measures (See Table 3.4 – Page 81).

3.7.4 Results of individual outcomes

A description of the different outcome measures is presented below, arranged in alphabetical order. All outcome measures investigated the burden on caregivers when caring for children with different long-term health conditions. Summaries of the methodological ratings, quality of psychometrics and best level of synthesis are presented in Table 3.5, Table 3.6 and Table 3.7 respectively.

3.7.4.1 Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)

The APSP-POM was developed to measure the impact of caring for a child with perinatal stroke in Canadian parents and families. Three studies using the scale were available [220-222]. There was limited evidence for content validity, structural validity, reliability and internal consistency (IC). Item and scale content validity indices were not calculated; the amount and handling of missing items were not described; a sub-optimal sample (N=58) was utilized, and the retest conditions of the test-retest reliability test phase were not specified. There was moderate evidence for construct validity; all three studies were of fair methodological quality. However, the handling of missing responses and hypotheses were not clearly outlined.

3.7.4.2 Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)

The ACEND was designed to measure the impact of caring for children with neuromuscular diseases [223]. Three studies using the scale were retrieved [125,126,223]. Evidence for content and structural

validity were unknown as the methodologies were poor [223]. No theoretical framework was utilised in item generation, factor analyses were not performed, and a sub-optimal sample was used (N=60). There was moderate evidence for construct validity. Two of the studies were of fair methodological quality [125,126]; however, hypotheses were not specific, and psychometrics of comparator instruments were not provided. There was limited evidence for criterion validity, and the handling of missing responses was not specified [223].

3.7.4.3 Autism Parenting Stress Index (APSI)

The APSI measures the burden of caring for a child with autism [224]. Three studies using the APSI were retrieved [224-226]. There was limited evidence for IC and content validity [224]. The handling of missing responses and the content validation process was not clearly outlined. There was unknown evidence for structural validity and reliability, an inappropriate rotational method was used, and a small sample (n=18) was recruited for test-retest reliability [224]. There was moderate evidence for construct validity, and methodologies were of fair quality as hypotheses were not specified [225,226].

3.7.4.4 Caregiver Burden Index (CBI)

Developed in China, the CBI measures the burden of caring for a child with allergies [64]. It was also translated and validated in Turkish [227]. There was strong evidence for content validity [64]. Evidence for structural and cross-cultural validity was unknown as retrieved studies were of poor quality, an inappropriate rotational method was used [64], and confirmatory factor analysis was not performed [227]. Evidence for IC was moderate, both studies were of fair quality, and the handling of missing responses was not described. Evidence for reliability was limited, and a sub-optimal sample (N=38) was utilised [227].

3.7.4.5 Caregiver Burden Inventory (CBIn)

The CBIn was initially developed to measure the burden of caring for adults with functional limitations [228-230]. A single study using the CBIn was retrieved [231]. Evidence for construct validity was unknown, the psychometrics of comparator instrument were not provided, and sub-optimal statistical methods were applied [231].

3.7.4.6 Caregiver Burden Scale (CBS)

The CBS was initially developed to measure the burden of caring for adult patients with either stroke or dementia [232]. Four studies applying the CBS were retrieved [233-236]. There was unknown evidence for construct validity as studies were of poor methodology. The handling of missing data and

psychometrics of comparator instruments were not described. Further, two studies were underpowered [235,236].

3.7.4.7 Caregiver Difficulties Scale (CDS)

The CDS was developed to measure caregiver burden in Sri Lankan caregivers of children with CP and three studies using the scale were available [25,49,237]. There was strong evidence for content validity [25,237]. Evidence for IC was moderate, and methodologies were of fair quality, i.e. the handling of missing responses was not explained [25,237]. There was conflicting evidence for reliability, structural validity and construct validity as methodologies were of either poor [25,49] or fair quality [237]. For instance, results for intra-class correlation coefficient (ICC) were not supplied [25], an inappropriate factorial rotational method was utilised [25], no description was given for comparator instruments [25], and hypotheses were not succinct [25,49,237]. There was limited evidence for trans-cultural validity, the handling of missing responses and demographic characteristics of the pre-test sample were not adequately described [237].

3.7.4.8 Caregiver Strain Index (CSI)

Originally developed to measure the burden of caring for the elderly with chronic diseases [238], the CSI was applied in seven studies [12,28,239-243]. There was unknown evidence for IC as the only study that evaluated IC was of poor methodology [12]. There was moderate evidence construct validity as the methodologies of retrieved studies were either of fair [12,28,239,241,243] or good [240,242] quality. The handling of missing responses was only reported in two studies [240,242], and hypotheses were not clearly outlined in all studies.

3.7.4.9 Caregiver Strain Questionnaire (CSQ)

The CSQ was designed to measure the impact of caring for a child with severe emotional and behavioural problems [244]. Two studies using the scale were retrieved [245,246]. There was strong evidence for construct validity as both studies were of good methodological quality. However, the magnitude of correlations between constructs was not specified [245,246].

3.7.4.10 Family Burden Interview Schedule (FIBS)

The FIBS was designed to measure the impact of caring for adult patients with psychiatric conditions [247,248]. Two studies using the scale were retrieved [249,250]. There was conflicting evidence for construct validity. One study was of poor quality, and the psychometrics of comparator instruments was not provided [250]. The second study was of fair methodological quality, but the handling of missing items and comparator instruments were poorly described [249].

3.7.4.11 Family Impact of Childhood Disability (FICD)

Two variations in the original and revised version were retrieved [251,252].

- **FICD - Original version**

The FICD was designed to measure the impact of caring for a child with a developmental disability [251]. Six studies using the scale were retrieved [251,253-256]. There was conflicting evidence for structural validity as one study was of fair quality; the handling of missing responses and factor rotation were not described [257]. The second study was of poor quality as it used the varimax (orthogonal) rotational method which was inappropriate [251]. There was strong evidence for IC as retrieved studies were of fair [251] and excellent [257] quality respectively. There was moderate evidence for construct validity, and all studies were of fair quality, but the handling of missing responses and hypotheses was not clearly outlined [251,253-257].

- **FICD - revised version**

The FICD +4 is a shortened, and revised version of the FICD and two studies using the scale were retrieved [252,257]. There was moderate evidence for IC and structural validity, the retrieved studies were of good quality, but the number of missing responses was not described [257]. There was strong evidence for construct validity. The two studies were of good quality; however, hypotheses were not clearly stated [252,257].

3.7.4.12 Family Impact Questionnaire (FIQ)

The FIQ was designed to measure the impact of caring for children with externalising behaviours [258]. Seven studies using the scale were retrieved [259-265]. There was moderate evidence for construct validity, and five studies were of fair quality [260-263,265]. The handling of missing responses, psychometrics of comparator instruments and hypotheses were not explicitly described.

3.7.4.13 Fathers of Children with Developmental Challenges Questionnaire (FCDC)

The FCDC was designed to measure the impact of caring for a child with a developmental disability on fathers, and a single study was available [266]. There was unknown evidence for content and construct validity. Details of cognitive interviews were poorly described, content validity indices were not calculated, the hypothesis was not specific, and psychometrics of comparator instruments were not provided. Evidence for IC and structural validity was limited. The sample was sub-optimal (N=85) and the handling of missing responses was not described [266].

3.7.4.14 Impact on Family Scale (IPFAM)

Two versions in the original [267] and simplified [268] versions of the IPFAM were retrieved.

- **IPFAM - original version**

The IPFAM is a 21-item tool which was originally developed to measure the impact of caring for children with chronic illnesses [267]. Fifteen studies utilising the scale were retrieved [126,269-282]. There was conflicting evidence for construct validity as studies were of poor [270,276-280,283], fair [269,271,275,281,282] and good [126,272-274] methodological quality. A lack of clear hypotheses, small sample sizes and no description of the psychometrics of comparator outcome measures were the major methodological shortcomings.

- **IPFAM - simplified version**

This is a 15-item, shortened version of the IPFAM [268]. Three studies using the tool were retrieved [268,284,285]. Evidence for IC and structural validity was limited as the retrieved studies were of fair quality. The handling of missing responses was not reported [268]. Evidence of construct validity was moderate, but the hypotheses and psychometrics of comparator outcome measures were not clearly outlined [284,285].

3.7.4.15 Maslach Burnout Inventory (MBI)

The MBI was initially developed to measure occupational burnout [223,286]. Four studies utilising the measure were retrieved [115,287-289]. Evidence for construct validity was unknown, and three of the studies were of poor methodological quality [115,287,288]. Hypothesis was not specific; the psychometrics of comparator instruments and handling of missing responses were not provided with one of the studies utilising a very small sample (n=18) [287].

3.7.4.16 Modified Caregiver Strain Index (M-CSI)

The M-CSI was validated initially in a geriatric population [290]. Three studies applying the MCSI were retrieved [89,90,291]. Evidence for construct (divergent) validity was unknown, and all three studies consisted of poor methodologies [89,90,291]. There was no description of psychometrics of comparator instruments [89,291], and a sub-optimal sample (N=25) was used in one of the studies [90].

3.7.4.17 Nijmeegse Ouderlijke Stress Index (NOSI)

The NOSI is the Dutch version of the Parenting Stress Index (PSI), and a single study was retrieved [292]. There was limited evidence for construct validity; the study was of fair methodological quality; handling of missing responses was not described, and hypotheses were not specific.

3.7.4.18 Parental Stress Scale (PSS)

The PSS was developed to measure the challenges associated with parenting [293]. Three studies using the scale were retrieved [294-296]. There was moderate evidence for construct validity as studies were of fair methodological quality. The handling of missing responses, hypotheses and psychometrics of comparator instruments were not clearly defined/outlined.

3.7.4.19 Parents of Children with Disabilities Inventory (PCDI)

The PCDI was developed to measure the burden of caring for a child with physical disabilities and two studies using the scale were retrieved [297,298]. There was substantial evidence for content validity. Evidence for IC and structural validity was unknown as the retrieved studies were of poor quality and factor analysis was not performed. Evidence for reliability was limited as a sub-optimal sample (N=31) was utilised [297]. Evidence for construct validity was moderate as the studies were of fair methodological quality [297,298]. Hypotheses and the handling of missing responses were not clearly outlined.

3.7.4.20 Perceived Stress Scale (PSS)

The PSS is a 14-item outcome measure originally validated to measure the impact of stressful life events [299]. Three variants in the PSS-4 [300,301], PSS-10, [299] and PSS-14 were retrieved.

- **PSS-4**

The PSS-4 is a four-item, brief version of the PSS-14 [300,301]. We retrieved four studies utilising the scale [302-305]. There was moderate evidence for construct validity, the handling of missing responses was only described in a single study [305], and hypotheses and psychometrics of comparator instruments were not clearly outlined in all four studies.

- **PSS-10**

The PSS-10 is a 10-item, revised version of the PSS-14. We retrieved seven studies utilising the scale [306-312]. There was moderate evidence for construct validity, but the handling of missing responses, hypotheses and psychometrics of comparator instruments were not clearly outlined in most studies.

- **PSS-14**

Ten studies using the scale were retrieved [16,301,313-320]. There was moderate evidence for construct validity, the handling of missing responses, hypotheses and psychometrics of comparator instruments were not explicitly outlined in most studies.

3.7.4.21 Parenting Stress Index (PSI)

The PSI was initially developed to measure the stress associated with parenting [321]. Two variants in the long (120-items) and short versions (36-items) were retrieved.

- **Parenting Stress Index - Long Form (PSI-LF)**

Nine studies were retrieved [322-330]. Evidence for trans-cultural validity was unknown as the adaptation methodology was poor [329]. There was moderate evidence for construct validity, but hypotheses and psychometric properties of comparator instruments were not clearly stated in most of the studies [322-325,327,328,330].

- **Parenting Stress Index-Short Form (PSI-SF)**

The PSI-SF is a 36-item shortened version of the PSI. Seventy-six (76) studies utilizing the measure were retrieved [17,251,257,261, 269,273,279,331-399]. There was unknown evidence for cross-cultural validity, the independence of translations was questionable, and CFA was not performed for structural validity evaluation [398]. There was conflicting evidence for structural validity as retrieved studies were of poor [398] and fair [321] quality; an inappropriate rotation method (varimax) was used, and handling of missing responses was not reported respectively. There was limited evidence for IC, the retrieved studies were of fair quality, and the handling of missing responses was not reported. There was moderate evidence for construct validity, most of the studies were of fair quality. Hypotheses, handling of missing responses and psychometrics of comparator instruments were not clearly outlined in most of the studies.

3.7.4.22 Questionnaire on Resources and Stress - Short Form (QRS-SF)

The QRS-SF [400] is a 52-item modification of the original QRS (285-items) [401] which was developed to measure the impact of caring for a child with a disability. 29 studies utilising the QRS-SF were retrieved [332, 402-430]. Evidence for cross-cultural and structural validity was unknown. Studies were of poor quality, i.e. in one study, factor analysis (FA) was not performed [429], and both orthogonal and oblique rotations were applied during FA in the second study [430]. There was conflicting evidence for construct validity, and there was an almost equal number of studies of either poor [402,404-406,409,411,412,415,420,423,428] or fair methodological quality [403,407,408,410,413,416-418,422,424,426,427,429,430].

3.7.4.23 Revised Burden Measure (RBM)

The RBM is a revised version of the burden scale which was originally developed to measure the impact of caring for the elderly [431]. Seven studies utilizing the RBM were retrieved [104,117,432-

436]. There was strong evidence for construct validity as three of the studies were of good quality [104,433,434]; however, hypotheses and psychometrics of comparator instruments were not clearly outlined.

3.7.4.24 Stress Level of Mothers with Children with CP Measurement Tool (SMCP)

The SMCP measures the stress associated with caring for a child with CP, and we retrieved a single study using the tool [136]. Evidence of construct validity was unknown as the study was of poor methodological quality. The handling of missing responses and hypotheses were not explicitly stated. Further, the adaptation of the comparator instrument was poorly done [136].

3.7.4.25 The Pediatric Quality of Life Inventory-Generic Core Scales (PedsQL)- Family Impact Module

The PedsQL is a validated measure which quantifies the effects of caregiving on informal caregivers' HRQoL and family functioning [437-440]. Eight studies utilising the scale were retrieved [278,441-447]. There was conflicting evidence for IC, and cross-cultural validity as retrieved studies were of poor [446] and good quality [445]. Confirmatory factor analysis was not performed, and the handling of missing responses was not documented. There was moderate evidence for structural validity as the handling of missing responses was not outlined [445]. Evidence for reliability was limited as a sub-optimal sample (N=47) was utilised [446]. There was moderate evidence for construct validity; most studies were of fair quality and hypotheses were not explicitly specified [441,443,445,447].

3.7.4.26 Zarit Burden Inventory (ZBI)

The ZBI was originally developed to measure the burden of caring for geriatric patients with dementia [448]. We retrieved six studies using the scale [449-454]. There was moderate evidence for construct validity as five of the studies were of fair methodological quality [449-451,453,454]. Hypotheses, psychometrics of comparator instruments and the handling of missing responses were not clearly outlined.

3.7.5 Items identified

In total, 136 items were identified from 32 different scales. (See Appendix 11.2 – Page 270)

3.8 Discussion

3.8.1 Settings

Most of the outcomes [29/31] were developed in high-income settings, especially in the USA [20/31]. Critical appraisal of both face and content validity of the outcomes is essential if identified outcomes are to be utilised in lower- and middle-income countries with different socio-economic and cultural contexts. Caregiver burden is a subjective and context-specific outcome [29,42,211, 212].

3.8.2 Content validity

Content validity is defined as the extent to which items on an outcome measure adequately represent the construct [455,456]. Ideally, there is a need for an amalgamation of findings from systematic review(s), panel of experts and target users of the patient-reported outcome (PROM) to develop a context-specific and culturally appropriate PROM [92,457-461]. Only three outcomes retrieved had sufficient evidence of content validation, and this could be a potential shortcoming. Content validity is envisaged as the "most important" psychometric property as lack thereof affects the quality of all other psychometric properties [456]. Further, most of the outcomes [23/31] were generic, and some [11/31] had been originally developed to measure burden in caring for adults. To ensure content validity, there is a great need to adapt items using robust methodologies to ensure that PROM is contextually relevant for use in a paediatric population [462-465]. For example, the item *"It is upsetting to find the person I care for has changed so much from his/her former self (For example: he/she is a different person than he/she used to be)……"* on the MCSI [290] may require adaptation for it to be applicable for use in caregivers of children with long-term health conditions. The item was presumably used as is in all the studies which utilised the MCSI. Rephrasing of the item may not be necessarily adequate; other psychometric analytical techniques such as factor analysis are essential to ensure that the adapted tool retains its content and structural validity [458,466-468].

3.8.3 Structural validity

Structural/factorial validity is defined as the extent to which scores on an outcome measure adequately reflect the dimensions/structure of the construct to be measured [455]. Only 11 outcomes retrieved were evaluated for structural validity. Further, of these, only two had moderate evidence of structural validity. Methodological shortcomings were in reporting of the amount and handling of missing values [220,237,268,321], sub-optimal sampling [266], no description of rotation method [257] and factor analysis was not performed in some studies [297,429]. Furthermore, some studies [25,64,224,227,251,398,430], utilized orthogonal rotation which is inappropriate when evaluating the

structural validity for PROMs measuring latent constructs [2,155-157,468]. Orthogonal rotation is used when factors are hypothesised to be unrelated [155,157,469], which is not the case for caregiver burden as factors are more likely to be interlinked as defined by most conceptual frameworks [209,284,331,342,419,433,470.472]. The lack of structural validity is of concern as it negatively affects the quality of other psychometrics such as reliability, responsiveness, construct and criterion validity [151-157]. Further, most retrieved outcomes [10/11] were tested using classical theory testing methods (CTT), i.e. factor analysis for structural validity evaluation. This could be a potential shortcoming for the overall quality of evidence for structural validity given the ever-increasing recognition of the "superiority" of item response theory (IRT) techniques such as Rasch analysis in dimensionality testing [159,160,473-476]. There is also a growing impetus towards complementary analysis whereby both CTT and IRT methods are applied concurrently for structural validity assessment [159-163].

3.8.4 Reliability

Reliability is the degree to which the measurement is free from measurement error and can be measured using various indicators [147,455]. Internal consistency (IC), which measures the extent of correlation between items with an outcome measure [455], was cited as the sole indicator of reliability across outcomes. Further, only a single outcome had moderate evidence of reliability [64,227]. Under CTT, the reliability of a test is a function of test length in accordance with Spearman-Brown's equation of prophecy [473,476,477]. Simply put, a tool is likely to be reliable if it contains more items, i.e. an outcome can yield high alphas even if there is inadequate evidence of structural validity [160,161,473]. For example, evidence for structural validity for the CBI was poor as incorrect rotation method was utilised; however, the test yielded a Cronbach alpha of 0.98 which would be considered as excellent evidence of reliability [64]. This invariably illustrates the criticism of the Cronbach alpha as a sole indicator of scale reliability. Other indices such as the alternative forms, split-half and test-retest reliability are recommended for concrete evidence of the reliability of outcome measures [154,158] as a tool can be reliable yet not valid [478]. More so, it is argued that, despite its wide usage and popularity, the Cronbach alpha is the least desirable index for reliability estimation [158,474]. According to Linacre (1997), Cronbach alpha overtly over-estimates the amount of test reliability as it is calculated from raw/unstandardized data/scores [457].

3.8.4.1 Construct validity

Construct validity refers to the extent to which scores on an instrument relate to other measures in a manner that is consistent with theoretically derived hypotheses concerning the concepts that are

being measured [455]. It was the most evaluated psychometric property. Depression, self-efficacy, anxiety and social support were amongst a plethora of outcomes against which caregiver burden was contrasted. Retrieved studies were of varying methodological quality, with only a minority [3/31] of outcomes displaying strong evidence of construct validity [245,251,431]. Most studies did not report the handling of missing responses, succinctly outline hypotheses, nor adequately describe the utility and psychometric properties of comparator instruments. These are crucial methodological quality benchmarks for construct validation evaluation [147,455]. Nonetheless, these findings were unsurprising as psychometric evaluation was not the primary aim for most studies analysed. The caveat is the tendency for "snow-ball referencing" in literature whereby authors tend to cite previously published studies to demonstrate/justify construct validity of outcome measures. This, therefore, underscores the importance of robust methodologies in the evaluation of construct validity regardless of study aims.

3.8.4.2 Criterion validity

Criterion validity is defined as the extent to which scores on an outcome measure performance against an established gold standard [147,455]. There was limited evidence for criterion validity as it was assessed only in a single study which was of fair quality [223]. Caregiver burden is a subjective, multidimensional, and a context-specific latent variable [29,42,211,212]; as such, it is difficult to establish a "gold standard" against which criterion validity can be assessed.

3.8.4.3 Clinical and research utility

In addition to psychometrics, other factors such as test length, mode of administration, cost and availability of scoring instructions and criterial values are important attributes when selecting an outcome for research and/or clinical usage [461,479,480]. To reduce respondent burden and subsequently reduce the amount of missing data, an "ideal PROM" should be acceptable to respondents, short, easy to administer, valid and reliable [41,480]. Most of the outcomes had 10-26 items [23/31], were rated on a 5-point Likert scale [21/31] and had scoring instructions available [19/31]. However, only a few items had information on cost [6/31], recall period [6/31] and approximate completion time [10/31]. Additionally, most outcomes [27/31] did not preset criterical values which make interpretation and comparison of results challenging, thus negatively affecting the utility of most identified outcome measures.

3.9 Items identified

These items were added to the item bank for inclusion in the new ZCCS and will be discussed in Chapter Five.

3.10 Limitations of the systematic review

The stringent nature of COSMIN as testified by almost similar reviews [481,482] could have been a potential limitation to the present review. Further, the COSMIN guidelines came into effect in 2011 whereas most of the outcomes [25/31] were published before 2012 and this could have contributed to poor methodological ratings for most outcomes. More so, psychometric evaluation was not the primary aim for most studies, and this might have affected the methodological quality ratings for most studies.

3.11 Conclusion

We identified 32 outcome measures, and most of the tools identified were generic and were not adequately adapted for usage in caregivers of children with paediatric disabilities/long-term health conditions. Construct validity was the most assessed psychometric property which underscores the need for evaluation of other psychometrics such as responsiveness and test-retest reliability. Concurrently, there is an expressed need for the establishment of criteria values, manuals for scoring and interpretation of scores. Psychometric techniques such as latent class analysis (LCA) are essential given the paucity of gold standards in evaluating criterion validity in latent constructs. Given that most outcomes were evaluated using CCT methods, there is also a need for application of contemporary psychometric methods such as IRT to develop outcomes with true metric properties. Where appropriate, there is a need for utilization of robust methodologies in either developing or adaptation of caregiver burden outcomes as most of the identified tools had limited psychometric properties. Further, clinicians need to critically appraise the utility of the appraised outcomes before the selection of an appropriate tool for use in routine patient care.

3.12 Tables

Table 3.4 : Caregiver burden outcome measures utility

Outcome	Country	Classification	Population - condition(s)	Year published	Completion Time	Response categories	Number of items	Score range	Criterial values	Scoring manual	Recall period	Cost
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM	Canada	Condition-specific	Paediatrics - Perinatal stroke	2014	5 minutes	5	26	1 - 86	none			
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)	USA	Condition-specific	Paediatrics- Neuromuscular disease	2011		5 & 6		41-228	none			
Autism Parenting Stress Index (APSI)	USA	Condition-specific	Paediatrics - Autism	2012		5	13	13-65	none			
Caregiver Burden Index (CBI)	Taiwan	Condition-specific	Paediatrics - Allergies	2014		5	20	20-100	none		2 weeks	
Caregiver Burden Inventory (CBIn)	-	Generic	Adults- functional limitations	1989	10-15 minutes	5	24	0-20	available	Available		
Caregiver Burden Scale (CBS)	Sweden	Generic	Adults- stroke & dementia	1996		4	22	22-88		Available		
Caregiver Difficulties Scale (CDS)	Sri Lanka	Condition-specific	Paediatrics- CP	2013	5-10 minutes	5	25	0-100	none	Available		Free

Caregiver Strain Index (CSI)	USA	Generic	Geriatrics	1983	2-5 minutes	2	13	0-13	available	Available		Free
Caregiver Strain Questionnaire (CSQ)	USA	Condition-specific	Paediatrics-behavioural problems	1997		5	21	21-105	none	Available	6 months	
Family Burden Interview Schedule (FIBS)	India	Generic	Adults-psychiatric conditions	1981	~60 minutes	5	56	56-280				
Family Impact of Childhood Disability (FICD)- original version	Canada	Generic	Paediatrics - disability	2002		4	15	10-40				
Family Impact of Childhood Disability (FICD)- revised version	France Denmark Ireland	Condition-specific	paediatrics- CP	2012		4	20					
Family Impact Questionnaire (FIQ)	USA	Generic	Paediatrics	1993		4	50	0-150	none			
Fathers of Children with Developmental Challenges Questionnaire (FCDC)	USA	Generic	Paediatrics-Developmental delay	2014		5	20	20-100	none	Available		
Impact on Family Scale (IPFAM)-original version	USA	Generic	Paediatrics-chronic conditions	1980		4	21		none	Available		

Impact on Family Scale (IPFAM)-simplified version	USA	Generic	Paediatrics-chronic conditions	2003	10 minutes	4	15		none	Available		
Maslach Burnout Inventory (MBI)	USA	Generic	Adults-General population	1981	10-15 minutes	5	22	0-88	none	Available		
Modified Caregiver Strain Index (M-CSI)	USA	Generic	Geriatrics	2003		3	13	0-26	none			Free
Nijmeegse Ouderlijke Stress Index (NOSI)	Nether-lands	Generic	Paediatrics	1992		6	25	25-150	none	Available		
Parental Stress Scale (PSS)	USA	Generic	Paediatrics-general	1995		5	18	18-90	none	Available		
Parents of Children with Disabilities Inventory (PCDI)	USA	Generic	Paediatrics- CP & spina bifida	1995		6	40	40-240	none			
Perceived Stress Scale (PSS-4)	USA	Generic	Adults - community-dwelling adults	1988		5	4	0-16	none	Available	1 week	
Perceived Stress Scale (PSS-10)	USA	Generic	Adults - community-dwelling adults	1988		5	10	0-40	none	Available	1 week	
Perceived Stress Scale (PSS-14)	USA	Generic	Adults - community dwelling adults	1988		5	14	0-56	none	Available	1 week	

PSI- Long Form (PSI-LF)	USA	Generic	Paediatrics-TDC	1976	~30 minutes	5	120		none	Available		Paid for
PSI-Short Form (PSI-SF)	USA	Generic	Paediatrics-TDC	1995	10 minutes	5	36		available	Available		Paid for
Questionnaire on Resources and Stress-Short Form (QRS-SF)	USA	Generic	Paediatrics	1982		5	52	52-260	none	Available		
Revised Burden Measure (RBM)	Portugal	Generic	Geriatrics	2006		5	16	16-80	none			
Stress Level of Mothers with Children with CP Measurement Tool (SMCP)	South Korea	Condition-specific	Paediatrics - CP	1997		5	44	44-220	none			
The Pediatric Quality of Life Inventory-Generic Core Scales (PedsQL)- Family Impact Module	USA	Generic	Paediatrics - chronic conditions	1999	4-6 minutes	5	36	0-100	none	Available	30 days	Paid for
Zarit Burden Inventory (ZBI)	USA	Generic	Geriatrics - dementia	1980		5	22	0-88	available	Available		

Table 3.5: Methodological ratings of retrieved caregiver burden outcomes

Tool	Internal consistency	Cross-cultural validity	Reproducibility-reliability	Content validity	Structural Validity	Construct validity – convergent	Construct validity- divergent	Criterion validity
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Fair [220]		Fair [220]	Fair [220]	Fair [220]	Fair [220-222]	Fair [220-222]	
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)								
Autism Parenting Stress Index (APSI)	Fair [224]		Poor [224]	Fair [224]	Poor [224]		Fair [225,226]	
Caregiver Burden Index (CBI)	Fair [64,227]	Poor [227]	Fair [227]	Excellent [64]	Poor [64,227]			
Caregiver Burden Inventory (CBIn)						Poor [231]		
Caregiver burden scale (CBS)							Fair [34b,34c] Poor [34a]	
Caregiver Difficulties Scale (CDS)	Fair [25,237]	Fair [237]	Poor [25] Fair [237]	Fair [25] Excellent [237]	Poor [25] Fair [237]	Poor [25] [49] Fair [237]		

Caregiver strain index (CSI)	Poor [12]					Fair [12,28,239,241,243] Good [240,242]		
Caregiver Strain Questionnaire (CSQ)						Good [245,246]	Good [245,246]	
Family Burden Interview Schedule (FIBS)							Poor [250] Fair [249]	
Family Impact of Childhood Disability (FICD)- original version	Fair [251] Excellent [257]				Poor [251] Fair [257]	Fair [251,253-257]	Fair [251,253-257]	
Family Impact of Childhood Disability (FICD)- revised version	Good [251]				Good [251]	Good [251,252]	Good [251,252]	
Family Impact Questionnaire (FIQ)						Fair [261]	Poor [259,264] Fair [260-263,265]	
Fathers of Children with Developmental Challenges (FCDC) Questionnaire	Fair [266]			Poor [266]	Fair [266]	Poor [266]	Poor [266]	
Impact on Family Scale (IPFAM)						Poor [276,278-280,283] Fair [269,275] Good [272-274]	Fair [269,271,275,281,282] Poor [270,276-280,283] Good [126,272-274]	

Impact on Family Scale (IPFAM)-original version						Poor [270,276-280,283] Fair [269,271,275,281,282] Good [126,272-274]		
Maslach Burnout Inventory (MBI)							Poor [115,287,288] Fair [289]	
Modified Caregiver Strain Index (M-CSI)							Poor [89,90,291]	
Nijmeegse Ouderlijke Stress Index (NOSI)							Fair [292]	
Parental Stress Index -long form (PSI-LF)		Poor [329]				Poor [326,329] Fair [322-325,327,328,330]		
Parental Stress Index -short form (PSI-SF)	Fair [351]	Poor [341]			Poor [398] Fair [351]	Poor [17,269,279,352,357,358,363,374,380,381,384,392,394,395] Fair [251,261,273,331-336,338-350,353-356,359-,364-373,375,376,378,379,382,383,385-391,393,396,397,399] Good [257,337,377,389]	Poor Fair [333,335,362]	
Parental Stress Scale (PSS)						Fair [294-295]		

Parents of Children with Disabilities Inventory (PCDI)	Poor [297]		Fair [297]	Excellent [297]	Poor [297]	Fair [297,298]	Fair [297,298]	
PedsQL- Family Impact Module	Poor [446] Good [445]	Poor [446] Good [445]	Fair [446]		Good [445]	Poor [278,442,444] Fair [441,443,447]	Poor [442,444,446] Fair [441,443,447] Good [445]	
Perceived Stress Scale-10 (PSS-10)						Poor [307,311] Fair [306,308-310,312]		
Perceived Stress Scale-14 (PSS-14)						Poor [36,301,315,319] Fair [294,313,314,316-318,320]		
Perceived Stress Scale-4 (PSS-4)						Fair [302-305]	Poor [302] Fair [303] Good [304,305]	
Questionnaire on Resources and Stress- Short (QRS-SF)	Fair [430]	Poor [429]			Poor [430]	Poor [402,404-406,411,415] Fair [403,407,408,410,413,418,419,424,425,429,483] Good [414]	Poor [402,405,406,409,411,412,415,420,423,428] Fair [403,407,408,410,413,416-418,421,422,424,426,427,429] Good [414]	

Revised Burden Measure (RBM)						Poor [117,435] Fair [432,436] Good [104,433,434]	Poor [435] Fair [436]	
Simplified version of the IPFAM	Fair [268]				Fair [268]	Fair [268,285]	Fair [268] Good [284]	
Stress Level of Mothers with Children with CP Measurement Tool (SMCP)							Poor [136]	
Zarit Burden Inventory (ZBI)						Fair [449-452]	Fair [449,450,452]	

Table 3.6: Ratings of quality of psychometric properties of retrieved caregiver burden outcomes

Tool	Internal consistency	Cross-cultural validity	Reproducibility- reliability	Content validity	Structural Validity	Construct validity – convergent	Construct validity- divergent	Criterion validity
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	+ [220]		? [220]	+ [220]	+ [220]	? [220,221] + [222]	? [220,221] + [222]	
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)				? [223]	? [223]		? [125,126,223]	? [223]
Autism Parenting Stress Index (APSI)	? [224]		? [224]	? [224]	? [224]		? [225,226]	
Caregiver Burden Index (CBI)	+ [64,227]	? [227]	+ [227]	+ [64]	? [64,227]			
Caregiver Burden Inventory (CBIn)						? [231]		
Caregiver burden scale							? [233-236]	
Caregiver Difficulties Scale (CDS)	? [25] + [237]	? [237]	? [25] + [237]	+ [25] [237]	? [25] + [237]	? [25,49,237]		
Caregiver strain index (CSI)	? [12]					? [12,28,239-242]		
Caregiver Strain Questionnaire (CSQ)						+ [245,246]	+ [245,246]	
Family Burden Interview Schedule (FIBS)							? [249,250]	

Family Impact of Childhood Disability (FICD)- original	? [251] + [257]				? [251,257]	? [251,253-257]	? [251,253-257]	
Family Impact of Childhood Disability (FICD)- revised	+ [251]				+ [251]	? [251,252]	? [251,252]	
Family Impact Questionnaire (FIQ)							? [259-265]	
Fathers of Children with Developmental Challenges (FCDC) Questionnaire	? [266]			? [266]	? [266]	? [266]	? [266]	
Impact on Family Scale (IPFAM)						? [269,272-,278-280,283]	? [126,269-276,278,283]	
Maslach Burnout Inventory (MBI)							? [115,287-289]	
Modified Caregiver Strain Index (M-CSI)							? [89,90,291]	
Nijmeegse Ouderlijke Stress Index (NOSI)							? [292]	
The original version of the Impact on Family Scale (IPFAM)						? [126,269-276-282]		
Parental Stress Scale (PSS)						? [294-296]		
Parents of Children with Disabilities Inventory (PCDI)	? [297]		? [297]	+ [297]	? [297]	? [297,298]	? [297,298]	
PedsQL- Family Impact Module	+ [445] ? [446]	+ [445] ? [446]	? [446]		+ [445]	? [278,441-444,447]	? [441-447]	

Perceived Stress Scale-10						? [306-312]		
Perceived Stress Scale-14						? [313-3320]		
Perceived Stress Scale-4						? [302-305]		
PS1-LF		? [329]				? [322-329]		
PSI-SF	? [351]	? [341]			? [351,398]	? [17,251,257,261,269,273,279,331-350,352-399]	? [333,335,362]	
PSS-4							? [302-305]	
QRS-SF	? [430]	? [429]			? [430]	? [402-408,410,411,413-415,419,424,425,429,483]	? [402-417,420-424,426-430]	
Revised Burden Measure (RBM)						? [104,117,432-436]	? [117,435,436]	
Simplified version of the IDFAM	? [268]				? [268]	? [268,285]	? [268,284]	
Stress Level of Mothers with Children with CP Measurement Tool (SMCP)							? [136]	
Zarit Burden Inventory (ZBI)						? [449-452]	? [449,450,452]	

Table 3.7: Best evidence synthesis of the psychometric properties of retrieved caregiver burden outcomes

Tool	Internal consistency	Cross-cultural validity	Reproducibility-reliability	Content validity	Structural Validity	Construct validity – convergent	Construct validity-divergent	Criterion validity
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Limited (+)		Limited (-)	Limited (+)	Limited (+)	Moderate (- -)	Moderate (- -)	
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)								
Autism Parenting Stress Index (APSI)	Limited (-)		Unknown (?)	Limited (-)	Unknown (?)	Moderate (- -)		
Caregiver Burden Index (CBI)	Moderate (++)	Unknown (?)	Limited (+)	Strong (+++)		Unknown (?)		
Caregiver Burden Inventory (CBIn)						Unknown (?)		
Caregiver burden scale						Unknown (?)		
Caregiver Difficulties Scale (CDS)	Moderate (+ +)	Limited (-)	Conflicting (+/-)	Strong (---)	Conflicting (+/-)	Conflicting (+/-)		
Caregiver strain index (CSI)	Unknown (?)					Moderate (+ +)		
Caregiver Strain Questionnaire (CSQ)						Strong (+++)		

Family Burden Interview Schedule (FIBS)							Conflicting (+/-)	
Family Impact of Childhood Disability (FICD)- original version	Strong (---)				Conflicting (+/-)	Moderate (+ +)		
Family Impact of Childhood Disability (FICD)- revised version	Moderate (- -)				Moderate (- -)	Strong (+++)	Strong (+++)	
Family Impact Questionnaire (FIQ)						Moderate (+ +)		
Fathers of Children with Developmental Challenges (FCDC) Questionnaire	Limited (-)			Unknown (?)	Limited (-)	Unknown (?)	Unknown (?)	
Impact on Family Scale (IPFAM)						Conflicting (+/-)	Conflicting (+/-)	
Maslach Burnout Inventory (MBI)							Unknown (?)	
Modified Caregiver Strain Index (M-CSI)							Unknown (?)	
Nijmeegse Ouderlijke Stress Index (NOSI)						Limited (-)		
The original version of the Impact on Family Scale (IPFAM)						Conflicting (+/-)		
Parental Stress Scale (PSS)							Moderate (+ +)	

Parents of Children with Disabilities Inventory (PCDI)	Unknown (?)		Limited (-)	Strong (+ + +)	Unknown (?)	Moderate (+ +)	Moderate (+ +)	
PedsQL- Family Impact Module	Conflicting (-)	Conflicting (+/-)	Limited (-)		Moderate (- -)	Moderate (+ +)	Moderate (+ +)	
PSI-LF		Unknown (?)				Moderate (+ +)		
PSI-SF	Limited (-)	Unknown (?)			Conflicting (+/-)	Moderate (+ +)	Moderate (+ +)	
PSS-4						Moderate (+ +)		
PSS-10						Moderate (+ +)		
PSS-14						Moderate (+ +)		
QRS-SF		Unknown (?)			Unknown (?)	Conflicting (+/-)	Conflicting (+/-)	
Revised Burden Measure (RBM)						Strong (---)		
Simplified version of the IDFAM	Limited (-)				Limited (-)	Moderate (- -)	Moderate (- -)	
Stress Level of Mothers with Children with CP Measurement Tool (SMCP)							Unknown (?)	
Zarit Burden Inventory (ZBI)						Moderate (+ +)	Moderate (+ +)	

4 Chapter 4: Caregivers' semi-structured interviews

4.1 Introduction

The systematic review did not reveal any caregiver burden/strain instrument which met the requirements of psychometric robustness. There was, therefore, a need to develop a psychometrically sound instrument which would be relevant to the Zimbabwe context. The first stage of developing an appropriate instrument is to identify candidate items that had both face and content validity. As caregiver burden is context-specific, it was essential to explore the lived experiences of Zimbabwean caregivers qualitatively to identify possible candidate items. This was done through engagement with the primary stakeholders, the caregivers themselves, using a semi-structured qualitative research design.

4.2 Study Setting

Participants were recruited from Mvurwi District Hospital, which is in Mashonaland West province, and is situated 105 km from Harare, the capital city of Zimbabwe. The district hospital was purposively selected as it has an active paediatric rehabilitation outpatient clinic, this ensured access to large pool of potential participants to choose the study sample from. Urban participants were recruited from Harare Central Hospital' Children Rehabilitation Unit (CRU). The CRU is the largest paediatric rehabilitation unit in Zimbabwe, this also ensured access to adequate pool of caregivers.

4.3 Participants

Participants were purposively selected to capture caregivers' experiences across the continuum of care. We recruited primary (informal), adult caregivers (≥ 18 years) of children with CP. Caregivers were chosen to represent a diversity of place of residence (rural vs urban), literacy rate, SES, relationship to the child, duration of caregiving and child's functional level. These factors were deemed essential predictors to caregiver burden as identified by the start-of-art literature review (See Chapter Two). Further, we set to recruit 14 participants as this allowed representation of each vital characteristic. An equal number of participants from both rural and urban areas were selected as an exploratory approach was used for the present study. Further, more participants caring for more functionally-dependent children, and who reported fewer financial resources were recruited. These were deemed as "rich information sources" as they would have provided greater insights of the challenges associated with caring for a child with CP in a resource, constrained setting.

4.4 Study instruments

4.4.1 Demographic questionnaire

A demographic questionnaire was used to extract the following caregivers' details: age, SES, literacy level, educational level, relationship to child and the severity of the child's disability (See Appendix 11.23. After capturing caregivers' demographics, the PI utilised the gross motor classification system (GMFCS) to confirm the extent of the child's disability to ensure that there was an equal representation of caregivers providing care to children with different levels of functional impairments. The GMFCS is a psychometrically-robust, five-point ordinal measure for severity of CP with levels one and five being least and most severely affected individuals [484]. The GMFCS is one of the most used classification systems and has been previously used in the research setting [29].

4.4.2 Interview guide

4.4.2.1 Development

A self-designed interview guide was developed to explore caregivers' experiences. The conceptual frameworks identified in Chapter Two were utilized to develop themes for categorisation of the interview questions. Sample questions included: "What problems have you encountered when caring for your child?", "Has there been changes in your family expenses because of the needs of the child?". Questions were arranged from general to more specific to give room for participants to fully express themselves. The following probing questions were used to gain further clarity: "Can you please tell me more?" or "Can you please give examples?"

4.4.2.2 Pretesting

The interview guide was peer-reviewed by the co-supervisor and a fellow PhD student who is a qualitative researcher. Questions were rearranged from general to specific based on the feedback received. Thereafter, the PI piloted the schedule through use of role play by interviewing a fellow PhD student. Role play was necessary for the refinement of the principal investigator (PI)'s interview skills, as the PI is a predominantly quantitative researcher. Based on the interviewee feedback, it was deemed essential to engage a professional translator in the translation of the guide from English to Shona, as the interview schedule had been developed in English. The forward translation was necessary as the interviews were conducted in both English and Shona languages to accommodate participants' preferences. Afterwards, the PI pretested the schedule by interviewing two caregivers of children with CP to ensure the clarity, comprehensibility and appropriateness of the questions given the sensitive nature of the topic. The interviewed participants endorsed the relevancy of the question, and no further amendments were done to the interview protocol. Please refer to Appendix 11.3 for the full interview guide.

4.5 Procedure and data collection

Potential participants were identified from the hospital registry through the assistance of resident therapists. The resident therapists notified the potential participants of the study and, if interested, caregivers were given the information sheet and consent form package. The PI then contacted the participants and scheduled a day for the interview which coincided with the day they brought their children for scheduled therapy sessions for both urban- and rural-based caregivers. On the day of the interview, the PI utilised the teach-back approach in obtaining written consent to ensure that caregivers participated as autonomous agents. The researchers read out the information leaflet which outlined the study procedures and ethical considerations. Thereafter, caregivers were requested to synthesise the meaning of the constructs and to repeat the questions in their own words before responding. After obtaining consent, the researcher interviewed the caregivers in a private room using the predesigned interview schedule. The interviews lasted between 30 minutes to an hour.

4.6 Data analysis

4.6.1 Analysis plan and theoretical framework

Data were analysed using a deductive, thematic analysis framework. Thematic analysis *"is a method for identifying, analysing, organizing, describing, and reporting themes found within a dataset"* [485]. A deductive approach implies that researchers have a pre-specified paradigm of analysing the data [485] [486]. We chose the deductive approach as the semi-structured interviews were based on theoretical frameworks identified from the narrative literature review. We did not expect to elicit entirely new themes/information as opposed to the items extracted from the systematic and narrative review; instead the aims of this phase were to elicit context-specific caregiver challenges. A six-step data analysis plan was followed, i.e. data familiarisation, coding, search for themes, themes review, defining themes and write up [486-489].

4.6.2 Steps utilised in the theme identification

Firstly, all interviews were audio-recorded and transcribed verbatim by a research assistant. After that, a second research assistant independently back-translated the responses from Shona to English, after which the PI checked the accuracy of the translation. The PI and a second researcher independently read the transcripts to familiarise themselves with the data. Both researchers also noted/wrote down some of the probable themes emerging from the first readings. Thereafter, the researchers met (peer briefing), and discussed and developed a codebook which was to be used for searching for the themes. Afterwards, the PI and the second researcher independently searched for themes. Both researchers made use of sticky notes to classify similar codes/sub-themes and made some diagrammatic connections to various codes. The researchers also independently ascribed tentative theme names.

Finally, both researchers and the supervisors reviewed and finalised the themes identified through an iterative discussion process [485,490,491].

4.7 Ethical considerations

The study adhered to the Helsinki declaration and the following ethical principles were upheld:

4.7.1 Autonomy

Caregivers were treated as autonomous agents as they were requested to consent before participating voluntarily. Caregivers gave written consent in the presence of a witness.

4.7.2 Privacy and confidentiality

Interviews were held in a private room and data were de-identified through use of pseudo-names to preserve privacy and confidentiality.

4.7.3 Justice

Caregivers were also provided with snacks after the interviews to compensate for their time. Further, a pre-set criterion was utilized to ensure justice and equity across participants.

4.7.4 Referrals

The narration of caregiving experiences is a sensitive topic and caregivers were given time during the interview to express their emotions. Where appropriate, caregivers were referred for counselling and further management after the interview. Caregivers were also advised that the interview could be terminated or paused at any time without any penalty.

4.8 Results

4.8.1 Demographic characteristics

All caregivers were females, and an equal number of caregivers across the continuum of severity of CP, educational level, place of residence, and SES was recruited (Table 4.1).

Table 4.1 : Demographics of caregivers for semi-structured interviews

Variable	Attribute	Rural	Urban
Gender	Female	7	7
GMFCS Level of child under care	1	1	1
	2	1	1
	3	1	1
	4	2	2
	5	2	2

Relationship to child	Mother	4	5
	Aunt	1	1
	Grandmother	2	1
Level of education	None	2	1
	Primary	2	1
	Secondary	3	4
	Tertiary	0	1
Perceived financial adequacy	Very inadequate	2	3
	Inadequate	2	2
	Neutral	1	1
	Adequate	1	1
	Very adequate	1	0

N=14

4.8.2 Emerging themes

Six major themes emerged from the data, and these are presented in Figure 4-1 (Page 100). Themes are presented/arranged within the ICF framework, i.e. impairments are presented first followed by effects on participation, and finally, environmental/contextual factors influence on caregiver burden/challenges. The results are pooled together as the emergent themes were independent of participants' place of residence (rural vs. urban).

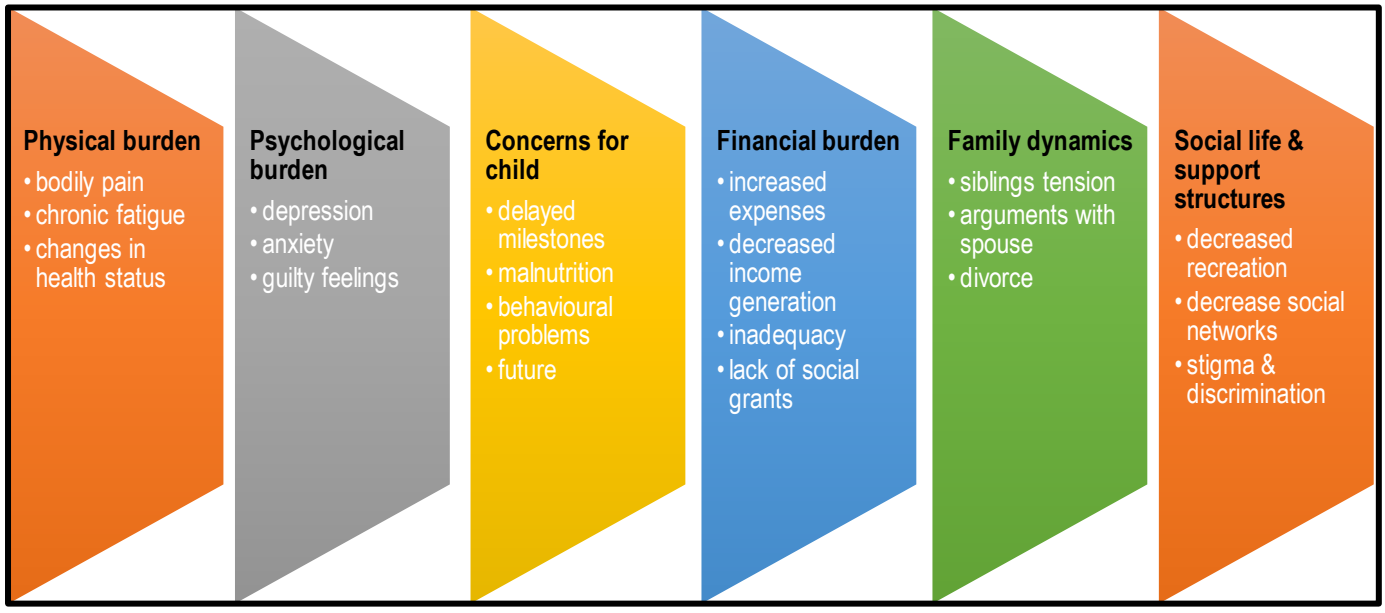


Figure 4-1: Themes from qualitative interviews

4.8.2.1 Physical burden

Caregivers reported changes in their overall physical health status with most caregivers (9/14) reporting bodily pain. Lifting and transfers contributed to joint and lower back pain; one caregiver said, *'…Y has become heavy, and I feel some pain whenever I lift her…".* Poverty worsened the physical burden as most caregivers could not afford aids and appliances. One caregiver said, *"…I cannot afford a wheelchair, and I have to carry my son on my back …"*. Lastly, caregivers also reported disrupted sleep, and this contributed to chronic fatigue.

4.8.2.2 Psychological concerns

Caregivers experienced poorer mental health due to the demands of caregiving. The notion of "kufungisisa", a local idiom for depression [492], evolved as a major sub-theme. Stigma and discrimination, lack of support structures and poverty were cited as the major sources of depression. One caregiver said, *"I can hardly sleep as I am always overwhelmed by the challenges I face every day in caring for my child…".* Caregivers also expressed anxiety over their child's future. Guilty feelings were also prevalent; caregivers either felt guilty for causing the child's condition or felt guilty for being unable to provide for the child fully. One caregiver said, *"I feel guilty as I am not able to be the parent I wish to be… I cannot provide C with all she requires…".*

4.8.2.3 Concerns for child

Most caregivers expressed concerns about the physical development of their children. One caregiver said, *"I am concerned that my child is not developing the way I expected compared to other children of the same age".* Some children had feeding problems and consequently were malnourished, and this was a source of concern for some caregivers. One caregiver said, *"I worry that my child is not gaining weight…she vomits almost anything that I feed her".* Caregivers were particularly concerned about the future of their child. *"I am worried if my child will be able to live independently as an adult…"* with another caregiver saying, *"I am anxious if my child will be able to find a suitable partner considering his disability…".* Caregivers also cited behavioural problems as major sources of distress, and this ranged from aggression, excessive crying, unpredictable sleep patterns to inappropriate urination. One caregiver said, *"I am worried that B is very aggressive and cannot play well with other children …. he often pinches others…".*

4.8.2.4 Financial concerns

Increased expenditure evolved as a central theme amongst all caregivers interviewed. Caregivers reported increased expenditure in specialist health services, aids and appliances, special diet and education services. One caregiver said, *"I am stressed that I cannot afford to provide a special diet for*

my child as she has problems in chewing and cannot eat the ordinary food we have as a family". Most caregivers (10/14) expressed that they either lost or down-scaled income generating opportunities upon the assumption of the caregiving role, and this contributed to the financial burden. One caregiver said, *"I am no longer able to engage in horticulture as much as I used to, and this was our major source of income…".* Caregivers also lamented the lack of social services by the government and this contributed to the financial burden. One caregiver said this: *"it is unfortunate that the government no-longer offer services to make our lives easier… I wish if they could offer us social grants like in other countries".*

4.8.2.5 Family dynamics

Caregivers expressed concerns about the alteration of family dynamics in the process of caring for a child with a disability. Caregivers reported "tension" erupting from siblings as most of the time finances were channelled towards the disabled child. One caregiver said, *"I feel guilty as I am not able to offer my children an equal amount of attention as X requires much attention as he cannot do anything on his own".* Some caregivers also reported having arguments with their spouses which were linked to the child's disability. In some instances, some caregivers reported marriage breakdown as one caregiver said, *"Our marriage ended in divorce as my husband did not want to take responsibility of raising our child because of her disability…".*

4.8.2.6 Social life and support structures

Caregivers expressed that they were overwhelmed by the caregiving roles and this negatively affected their social life. Some caregivers expressed that they could hardly find time to socialise or visit their family and relatives. One caregiver said this: *"… it is now hard for me to visit my relatives as it is challenging to move around with B … I last visited my relatives when she was still very young…"*.

Caregivers cited a lack of both formal and informal support structures as a stressor. Some caregivers also expressed a lack of empathy and support from some healthcare professionals. One caregiver was dissatisfied with the way she was treated, *"I wish that they (healthcare workers) would treat me with respect as much as they do for caregivers of healthy children".*

Stigma and discrimination also evolved as a sub-theme. Caregivers reported that their communities were not particularly accommodative and were not willing to assist. One caregiver said, *"I cannot even leave my child in the care of my neighbours as they believe that cerebral palsy is contagious…".* The stigma was also associated with myths whereby disability was linked with witchcraft. Four caregivers revealed that their communities could not embrace their children. One caregiver said, *"They cannot allow my child to play with their children as they believe that the disability was because of witchcraft".*

4.9 Discussion

The interviews confirmed that caring for a child with a disability is associated with caregiver burden as reported in the literature [83,493-495]. Caregiver burden evolved as a multi-dimensional construct as caregivers were affected emotionally, physically, socially and financially, representing the ICF constructs of impairments, activity limitations, participation restriction and contextual factors.

4.9.1 Physical burden

Caregiving is often associated with physical health problems such as headaches, hypertension, musculoskeletal disorders and peptic ulcers which require formal attention [50,83,144,312,496,497]. Unfortunately, caregivers residing in low-resource settings are not routinely screened for these ailments. As cited in previous studies, a lack of respite facilities, aids and appliances and special schools increases the risk of these ailments in caregivers thus leading to physical burden [493,494,498]. For example, several studies have shown that joint and low back pain arising from lifting and transfers is recurrent in CCWCP [18,83,124,239,499-501]. As reported elsewhere [18,83,124], most caregivers in the present study carried their children on their backs as they could not afford wheelchairs, and this was cited as the primary source of bodily pain. Unfortunately, the burden of musculoskeletal disorders is inter-linked with poverty which creates a vicious cycle which results in poor caregiver well-being.

4.9.2 Psychological burden

Caregivers overwhelmingly confirmed that the increasing demands of caring for a child with functional limitations lead to poor mental health [83,498]. As with previous qualitative studies, the presence of behavioural problems in children with CP, stigma, and worrying about the well-being of the child contributed to poor psychological functioning [495,498,502]. Diagnosis of CP can be considered as a "stressful life event", and in the absence of adequate support structures, some caregivers may fail to adjust to the demands of the caregiving role [495,498,502]. Although caregiving may have positive effects such as increased bonding and expanded social networks through caregiver support groups, studies have shown that the adverse effects invariably overshadow positive effects [2,5,8,40]. Caregivers are therefore prone to a myriad of feelings which range from hopelessness, anxiety, suicidal ideation, fear of the unknown, despair and guilt to feeling overwhelmed by the caregiving role [17-23,493,498,503,504]. This, therefore, underscores the need to provide counselling and psychological support to caregivers [23,495,493] as the lack of formal support for health-care services such as counselling for caregivers further compounds the caregiver burden [83,493,494]. Studies have shown that caregivers are likely to cope better if provided with information on the aetiology, prognosis and management of the condition [18,23].

4.9.3 Financial burden

Financial burden emerged as a key theme, and this was predictable given the current socio-economic turmoil in Zimbabwe [492,505,506]. For instance, 85-90% of Zimbabweans are unemployed, and more than 80% of the population survive on less than US$2 a day [505-507]. The lack of social grants and public health insurance further exacerbates the financial burden. Studies have shown financial burden to be prevalent even in caregivers of children with CP (CCWCP) residing in high-income countries who receive government social grants [136,494,508-511]. It was therefore unsurprising that all participants in the present study reported a financial burden. Against this general societal backdrop of financial deprivation, the caregivers are even further disadvantaged. The need for a special diet, frequent hospitalisations and decreased opportunities for income generation in a deteriorating economy partially accounted for the high burden. As children with CP in Africa are likely to present with multiple co-morbid conditions [23], this is likely to further exacerbate the financial burden due to the greater health-care expenditure. As in this study, poverty has been previously cited as a pivotal predictor of high caregiver burden in other low resourced settings such as Botswana, Malawi, Uganda and Kenya [23,493].

4.9.4 Family and social life

The excessive demands of caregiving can prejudice caregivers' opportunities for socialisation and recreational activities, leading to a psycho-social burden [498]. Stigma within communities further decreases caregivers' social networks and social participation [7,85,88-89,498]. Additionally, enacted stigma has also been reported as caregivers tend to seclude themselves from participation in social activities as a protective mechanism [498,502]. Caregivers in the present study attested that they were hesitant to freely bring out their children as they feared humiliation and stigmatisation in public circles.

Caring for a child with a physical disability can negatively affect family dynamics [83,107-109]. This was also confirmed in the present study as caregivers expressed guilt over failure to adequately attend to the needs of other family members. Further, if not properly handled, the distress can, unfortunately, lead to marriage breakdown. Divorces have been specially reported in low-resource settings where myths towards the aetiology of CP are still prevalent [86-89]. In high-income settings, there also have been reports of marriage tension and increased rate of conflicts due to the stress associated with caregiving [83]. This is unfortunate as social support, especially spousal support, is an essential buffer to caregiver burden [3,54,83,209,512-521] and may mitigate the financial burden related to caregiving.

4.9.5 Support structures

It is unfortunate that myths surrounding the causative agents of CP are still prevalent in Africa. For instance, the beliefs that CP is a result of witchcraft, bad luck and maternal promiscuity are still common [17-23,493,498,503,504]. The negative societal attitudes, unfortunately, result in caregiver distress and exclusion of CWCP in participation in day-to-day activities [7,23,85-89,124,498]. In extreme cases, in our African communities, caregivers are expelled from their communities, some children are locked up, and caregivers may perform filicide as communities are unlikely to accept children with CP [18,22,23,493. These results point to the need to further educate the community regarding the causes of disability and have locally based health or rehabilitation workers who can provide the support and education required.

4.10 Methodology critique

Study findings need to be interpreted with caution in the light of several methodological limitations. Firstly, the sample might not have been representative of the Zimbabwean caregivers of CWCP as participants were recruited from only two institutions. However, the study aimed to explore the lived experiences of caregivers qualitatively, and thus attainment of theoretical saturation as opposed to attaining external validity was more critical. Secondly, institution-based recruitment of participants could have resulted in selection bias as caregivers who present to hospitals may be coping better with the caregiving situation as studies have shown that caregivers faring worse are likely to lock up their children in the worst-case scenario [493]. Thirdly, all respondents were female, and this could potentially limit "generalizability"; however, in the African context, caregiving is considered a "feminine" role [12,522]. Nevertheless, fathers of children with disabilities are also likely to be affected, and future studies are needed to understand the impact of childhood disability on male caregivers.

Fourthly, solitary backward translation of scripts might have reduced the study rigour and trustworthiness. Ideally, transcripts ought to be independently translated by two or more people [523]. However, solitary translation is defensible in the context of "small q" thematic analysis [524] as the principal aim of the present study was to elicit items which had not have been captured through a prior systematic review. More so, the PI is proficient in both source and target languages, and this is important in the preservation of meaning. Although member and external checks were not performed, two researchers independently read all scripts before coding, and this might have increased the study trustworthiness [487,498]. Furthermore, the interview guide and analytical framework (code manual) were peer-reviewed and guided by the synthesis of previously validated conceptual frameworks, and findings from a systematic review and we believe this could have also

increased the study rigour and trustworthiness [48,98,105,107,108,525]. More so, the variability in the sample could have increased the credibility of our findings, i.e. we recruited caregivers of varying SES and who were caring for children with varying functional limitations. We predominantly utilised a deductive approach to analysis, and this can be envisaged as both a weakness and a strength [526,527]. It is a weakness as we may have potentially missed subtle themes not enshrined in the study conceptual framework [527,487]. On the contrary, we anticipated that the findings would fit within the context of previous studies. Caregiver burden is trans-cultural, and we did not expect caregivers' experiences to be orthogonal to other caregivers from different socio-cultural contexts.

4.11 Conclusion

Despite the methodological limitations [528], study findings confirmed some of the challenges faced by caregivers of children with disabilities residing in a low-resource setting. Deterioration in physical and psychological health, concerns for the child, financial concerns, alteration in family dynamics and social life and lack of support structures evolved as crucial challenges. Further, unique sub-themes such as challenges in finding suitable accommodation for urban dwellers, a lack of aids and appliances and cultural beliefs towards the aetiology of CP evolved as unique indicators when compared to the items gleaned from the earlier systematic review of published caregiver burden outcome measures. The information elicited was subsequently used to inform the development of a context-specific outcome measure to understand caregivers' experiences on a larger scale.

5 Chapter 5: Content validation of the Alpha version of the ZCCS

5.1 Introduction and objectives

It emerged from the systematic review of instruments that the existing caregiver burden scales were not all psychometrically sound. Additionally, the interviews with the caregivers indicated that there were certain factors which may contribute to strain which were unique to the Zimbabwean context. This chapter, therefore, describes the process of constructing an item bank, based on the review and the interviews, and then pruning the items through an iterative process.

5.2 Overview of the item reduction process

The first stage was to ensure that all relevant items were included. As there was duplication evident in the items gleaned from the systematic review, these were then combined into single items. Additionally, there were some items that were of little cultural relevance to the local population and these needed to be removed. This preliminary list was developed by the PI after pruning the comprehensive list of items identified through the systematic review and informed by the caregiver interviews. The list was pruned further through a series of consensus meetings with the co-supervisor, an occupational therapist with considerable experience in the field of childhood disability and a Shona speaker. The pruning was continued over two rounds of panel of experts input before being subjected to cognitive debriefing with the caregivers and finally psychometric testing (Table 5.1 Below).

Table 5.1: Process of ZCCS item reduction

Version	Activity	Items included
Initial item bank	Creation of initial item bank- items were extracted from measures identified through SR and caregivers' interviews	136
Version 1.0	The removal of redundant and inappropriate items by PI. Combining similar items into single questions	80
Version 2.0	Consensus meetings with co-supervisor (expert in the field of paediatric rehabilitation).	77
Version 3.0	Panel of experts Round 1	56
Version 4.0	Panel of experts Round 2	49
Version 5.0	Caregivers cognitive debriefing	45
Alpha version	Field testing	45

5.3 Development of Version 1.0 based on the systematic review, caregiver interviews and panel of experts

5.3.1 Introduction

The themes arising from the semi-structured interviews with the caregivers, and the paediatric caregiver burden conceptual framework by Raina et al. [48,98] were used as an initial schema within which to categorise the items. The themes included: physical burden, impact on self, economic/financial burden, impact on family, concerns for the child, social support, psychological burden, social life domain and overall perception of caregiver burden. The PI had extracted 136 candidate items from 31 tools identified from the SR and four (4) unique items from the caregivers' interviews. Each item was then categorised under one of the themes identified. Table 5.2 below is an illustration of how items under financial burden were extracted (See Appendix 11.2 - Page 270) for the rest of the domains).

Table 5.2: ZCCS financial burden item generation

Outcome Measure	Subtheme	Item
Caregiver difficulties Scale	Expenses	Is there an increase in your family expenses due to the child's condition?
Caregiver Burden Scale	Expenses	Spending a large amount of money
Caregiver difficulties Scale	Adequacy	Is your income adequate to provide the necessities for the child?
Zarit Burden Interview	Adequacy	Do you feel that you don't have enough money to take care of your child in addition to the rest of your expenses?
Caregiver Burden Scale	Adequacy	Our finances are not able to take care of other family members
Caregiver Strain Index	Work adjustments	There have been work adjustments (For example: I have to take time off for caregiving duties)
Caregiver Strain Questionnaire	Work adjustments	Missing work or neglecting other duties
Caregiver Strain Questionnaire	Other	Any family member having to do without things
Caregiver difficulties Scale	Other	Do you worry that you are unable to provide special facilities and services needed by your child?
Parenting Stress Index	Other	I am unhappy with the last purchase of clothing I made for myself
Caregiver Strain Index	Overall	Caregiving is a financial strain
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Financial burden	My child's condition places financial strain on my family

Before the consensus meetings with the co-supervisor, the PI further utilized thematic analysis for item reduction. Conceptually similar items, n=8 were merged into one item, e.g. items 47-50 were merged into one. Items were eliminated if they were not culturally appropriate (n=3), it was challenging to find semantic equivalency in the local language (n=2), not applicable to all children (n=2) and if the items were deemed to be non-specific (n=3). 18 items were deleted at this stage as summarized in Table 5.3 below .

Table 5.3: Amendments to the initial item list to develop version 1.0 of the ZCCS

Decision	Items	Motivation
Merged into one question as they all had a similar underlying meaning	e.g. Q47. Do you worry that your child gets insulted and ridiculed by others?	Although differently phrased, these four items share the same underlying conceptual meaning, i.e. stigma, thus the decision to combine them into one item
	Q48. I am worried about bringing my sick child out to meet other people?	
	Q49. Other people do not know how to treat my child?	
	Q50. Do you have to face embarrassing situations when you are travelling with the child?	
Merged into one question as they all had a similar underlying meaning	Q79. No one is to blame for my child's condition	Both items have the same underlying concept, acceptance of the child's disability, hence decision to merge
	Q83. I have come to terms with my child's condition	
Merged into one question as they all had a similar underlying meaning	Q91. Since having my child, I have been unable to try new and different things	Both items have the same underlying concept, changes in caregivers' lifestyle, hence decision to merge
	Q92. Since having my child, I feel that I am almost never able to do things that I like to do	
Eliminate as item not applicable to all children	Q54. When my child is actively playing, I find myself worried that s/he will get hurt.	Item may not be applicable to children with severe disabilities
Eliminate as items are culturally inappropriate	Q59. My child seems lonely.	Concepts of child's loneliness and expression of parental jealousy were deemed to be culturally distant to the target population hence the decision to eliminate them
	Q89. I am jealous of parents who have healthy children	
Eliminate as the item is too general	Q43. There are some things my child does that bother me a lot Q70. I find myself easily agitated	Both items are non-specific and are subject to different interpretations and would be potentially confusing to respondents
Eliminate as challenging to find semantic equivalency in the local language	Q56. Do you fear that your child will have accidents as a result of his/her disability?	Unclear to what "accidents" refer

5.3.2 Development of Version 2.0 after consensus meetings

The PI and co-supervisor qualitatively then analysed the second version of the ZCCS and reached consensus on which items to include. A further three items were dropped, and three items were rephrased to improve clarity (See Table 5.4 below). This second version was then used as a basis for the first panel of experts' content validation round one (See Appendix 11.4 Page 280).

Table 5.4: Amendments to ZCCS Version 1.0 to develop Version 2.0

Item	Action
I worry that my child is not developing physically as other children of his/her age? (For example, he/she is underweight)	Removed as the question is ambiguous and does not differentiate between growth and physical and/or mental development
In general, I often have the feeling that I cannot handle things very well	Removed as it was too non-specific
I find myself giving up more of my life to meet my child's needs than I ever expected	Removed as it was deemed redundant as almost similar to the item, "In general, I feel that my social life has suffered because of providing care to the child."
In general, do you feel that your social life has suffered because of providing care to your child?	Item was rephrased to change it from second-person to the first person; it was changed to "In general, I feel that my social life has suffered because of providing care to my child."
I worry that I am unable to provide special facilities and services needed by my child	Addition of an illustration: I worry that I am unable to provide special facilities and services needed by my child (For example, providing for a special diet)
The item "I feel isolated and alone as a result of caring for a child."	Was amended to "I feel isolated and alone/ (without friends) as a result of caring for the child" to improve clarity

5.4 Development of Version 3.0 and 4.0 after a panel of experts' content validation process

5.4.1 Introduction

A panel of experts examined the content validity of the third and fourth versions by rating the relevancy, clarity and appropriateness of each item. Panellists also rated the clarity of scoring instructions and layout of the ZCCS. The participants were drawn from different disciplines, and all had a post-graduate qualification, a minimum of eight years of clinical experience, and almost all had experience in paediatric neurology and prior experience with tool validation.

5.4.2 Recruitment and selection

The panel consisted of clinicians, and academics in the fields of rehabilitation, nursing, paediatrics, neurology, psychology, social work and psychiatry. Panellists were purposively selected to represent divergent views. The following selection criteria were used in selecting panellists: a minimum of five years of clinical experience, experience in working in paediatric neurology and mental health settings and being available and willing to participate in roundtable discussions. Eight panellists were recruited for each round, as the literature suggests that at least six (6) panellists are required [529-531]. We purposively selected three panellists who made extensive contributions in the first round of analysis for the second round. Further, five new panellists with diverse research and clinical backgrounds were recruited to gain as much input as possible. The characteristics of the panellists are presented in Table 5.5 (Page112). All experts were from Zimbabwe as it was important to develop a culturally appropriate outcome measure. As much as there is advocacy of recruitment of international subject experts [532], the initial item bank gleaned from the SR was reflective of a global understanding of caregiver experiences as most of the outcomes were developed in various high-income settings. It was thus clear that it was more important to have the input of local experts given the latency and cultural variations in the expressions of caregiving challenges [97,98].

Table 5.5: Panellist characteristics

Profession	Qualifications	Years of clinical experience	Experience in paediatric neurology mental health work setting	Prior experience with tool validation	Rounds involved
Occupational Therapist	PhD OT student; MSc OT	8	Yes	Yes	1
Physiotherapist	PhD PT; Msc Management	25	No	Yes	1
Physiotherapist	Msc. PT	35	Yes	No	1
Psychiatric Nurse	Msc. Psychiatric Nursing	9	Yes	Yes	1
Psychiatrist	M Med. Psychiatry	17	Yes	Yes	1
Clinical Psychologist	Msc. Clinical Psychology	13	Yes	Yes	1,2
Occupational Therapist	Msc. Public Health; Bsc (Hons) OT	11	Yes	No	1,2
Occupational Therapist	PhD, MSc Epidemiology	18	Yes	Yes	1, 2
Occupational Therapist	MSc OT, MSc Disability studies	30	Yes	Yes	2
Occupational Therapist (OT)	PhD OT student; MSc OT	9	Yes	Yes	2
Occupational Therapist (OT)	PhD OT; MSc Public Health	14	Yes	Yes	2
Physiotherapist	PhD PT student; MSc PT	27	Yes	No	2
Psychiatric Nurse	MSc. Psychiatric Nursing	15	Yes	Yes	2

N=8 first round, N=6 second round.

5.4.3 Instrumentation

The items in Version 2.0 were then formatted into a questionnaire (See Appendix 11.4 Page 280). Additionally, participants were also encouraged to provide additional comments as deemed necessary.

5.4.4 Procedure

Permission to carry out the study was granted by the University of Cape Town (Appendix 11.5 - Page 286) and the Medical Research Council of Zimbabwe (Appendix 11.6 - Page 287). The PI with the assistance of the co-supervisor afterwards drafted a list of potential panellists. Thereafter, after

applying the pre-set criterion, shortlisted personnel were physically approached by the PI who briefly explained the study concept. Participants who displayed interest and commitment to participate in the content validation process were then provided with a printed package containing an explanatory letter which outlined the concepts informing the development of the ZCCS, operational definitions and scoring instructions. Participants were requested to rate item relevancy on a four-point Likert scale independently, i.e. 1=not relevant, 2=somewhat relevant, 3=quite relevant and 4=highly relevant. Panellists also assessed the clarity of the items and were encouraged to express additional comments where necessary. Panellists were given up to two weeks to return the completed questionnaires. The PI sent email reminders after a week to increase the response rate and facilitate timely completion of the questionnaires. The results of the first round were analysed, and Version 3.0 was produced. This was then circulated to the Round two (2) participants who were requested to rate the content validity of the items. The same data collection procedures as with the first round were followed in collecting data in the second round.

5.4.5 Data management

Raw data from panellists were locked in a secure locker at the Department of Rehabilitation, the University of Zimbabwe and only the researcher had access to it. Digital copies of the data were encrypted and stored on a password-locked laptop and backed up on Dropbox.

5.4.6 Data analysis

Item content validity index (I-CVI) was calculated as a proportion of experts who rated either a 3 or 4 over the total number of panellists. Both the scale content validity index-average (S-CVI/Ave) and the scale content validity index-universal agreement index (S-CVI/UA) were calculated. The sum of the I-CVIs was divided by the total number of items to calculate the S-CVI/Ave, and S-CVI/UA was calculated by summing up all ratings of either 3 or 4 over the sum of panellists' ratings [529,533]. As with other consensus-based indices, the CVI is prone to inflated values due to an agreement by chance. A modified Kappa statistic (k*) which adjusts for agreement by chance was thus calculated. Items with an I-CVI ≥ of .78 and k* ≥.72 were selected, and the threshold for S-CVI was set at .9 as advised in the literature [529,533].

After Round 1 data were analysed, items which did not meet the criteria were excluded and the remaining items incorporated into Version 3.0. A similar analysis was performed on the responses to this version.

5.4.7 Ethical considerations

Panellists agreed to participate voluntarily upon signing of a consent form (See Appendix 11.7 – Page 288). Pseudo names were used to preserve identities of panellists. No responses were linked to specific participants.

5.5 Development of questionnaire layout and scoring instructions

The PI firstly reviewed the scoring options and scoring instructions of caregiver burden outcomes retrieved from the SR. After discussions with supervisors, it was agreed to use a five-point Likert scale with scoring options ranging from "strongly disagree=1" to "strongly agree=5". We also anticipated that some of the caregivers would have low literacy levels, so we, therefore, decided also to make use of pictorial prompts to increase the comprehensibility of the scoring instructions. The same strategy has been successfully used in validation studies in Uganda [3] and Malawi [47], countries with almost the same demographic profile. We also developed self-administered and interviewer-administered versions of the ZCCS to cater for caregivers with different literacy levels. For instance, the self-administered questionnaire was phrased in first-person pronouns, i.e. "I have …" whereas for the interviewer-administered version, questions were phrased in the second-person pronouns, i.e. "Have you …".

5.5.1 Results of panel of experts round 1

After analysis of the responses of the panellists, a further 21 items were removed based on a synthesis of panellists' ratings and suggestions (See Table 5.6 – Page 115). Items which were removed are flagged. The I-CVI median was 1.0 (range =.63-1.00). The S-CVI/Ave and S-CVI-UA were .913 and .912 respectively. Further, based on the recommendations by experts, the phrasing of the questions was also amended to reflect the first-person response, i.e. *"I experience…"* rather than *"Do you experience…?"*

Table 5.6: Items removed after Round 1

Question	I-CVI	k*	Decision	Comments
When I compare my general level of health over the past 12 months, I feel that my state of health today has worsened.	0.75	0.72	Omit	Redundant, similar to Q2, too generic, large recall period.
Having a child with a disability makes the decision to have more children difficult.	0.63	1.0	Omit	Low CVIs, may not be relevant to all groups - not discriminant enough
There have been changes in work plans due to caregiving (For example I had to turn down a job, or I cannot engage in farming activities as much as I used).	1.0	1.0	Omit	Redundant- similar concept as Q 12, combine with Q12
My income is adequate to provide the necessities for the child.	1.0	1.0	Omit	Redundant – info captured in Q14 Consult
My income is inadequate to take care of other family members.	0.63	0.63	Omit	Not directly related to caregiving burden
There have been adjustments/disruptions in family routines and social activities.	0.88	0.88	Omit	The same concept captured in social life questions, difficult to translate
I feel that caregiving has decreased our family standard of living, for example, some family members had to do without necessities.	0.75	0.72	Omit	Too generic, redundant as it is almost similar to Q16, not culturally appropriate
My child gets upset easily over the smallest thing.	0.88	0.88	Omit	Redundant- similar to Q27, difficult to translate
My child is not able to do as much as I expected.	0.88	0.88	Omit	Redundant – similar to Q35
I worry about something bad happening to my child when s/he is out of my care.	1	1	Retain	
I have to constantly wake up during the night and check on my child.	0.75	0.72	Omit	Redundant – similar to Q 5
I am unable to discuss your child's problems with other family members.	0.88	0.88	Omit	1.Redundant – combine with Q44 2. Item unclear
I expected to have closer and warmer feelings for my child than I do, and this bothers me.	0.63		Omit	Low ICV and not also culturally appropriate
In general, I often have the feeling that I cannot handle things very well.	0.75	0.72	Omit	Too generic and is not directly linked to the caregiving process
I worry that something I did, or my partner did during pregnancy caused my child's condition.	0.88	0.88	Omit	Combine with Q 57 as the concepts are almost similar

Seeing healthy children doing everyday activities makes me feel sad.	0.88	0.88	Omit	Redundant – similar to Q 35
I feel that I have lost control of my life because of caring for a child.	0.88	0.88	Omit	Redundant – similar to Q 5
I find myself to be easily agitated.	0.88	0.88	Omit	Too generic and not context specific to the caregiving situation
Since having my child, I have been unable to try new and different things.	0.75	0.72	Omit	Redundant- similar to Q70, not specific and not culturally appropriate
I feel trapped by my responsibilities as a parent/guardian.	0.88	0.88	Omit	Redundant, combine with Q72 as they are also similar constructs
I am unable to care for child much longer	0.75	0.75	Omit	Redundant and has a lower ICVI

5.5.2 Results of panel of experts' second round

Eleven items were further removed based on panellists' recommendation. Items deemed redundant (n=6), too generic (n=1) and not culturally appropriate (n=1) were dropped. Further, three pairs of almost conceptually equivalent items were merged, nine items were re-worded, and illustrations were added to two items to improve clarity (See Table 5.7 below).

Table 5.7: Item selection - panel of experts' second round

Domain	Sub-Theme	Question	I-ICV	K*	Decision & comments
Self	Time pressure	Considering my caregiving responsibilities, I do not have enough time for my basic needs such as having meals, bathing, etc.	1	1`	Retain
Self	Plans	Due to the responsibilities of caregiving, it is now difficult to plan for the future (for example, it is now difficult to plan to have other children).	0.88	0.88	Retain and add an illustration
Self	Health	In general, I feel that my health has suffered because of the care I provide to the child.	1	1`	Eliminate; too generic
Self	time pressure	I find myself giving up more of my life to meet my child's needs than I ever expected.	1	1`	Eliminate: not culturally appropriate
Physical	Sleep	My sleep is disturbed (for example the child I care for cries a lot and wakes me up at night).	1	1`	Retain
Physical	Exhaustion	I feel tired and exhausted because of caring for the child.	1	1`	Retain
Physical	Pain	I usually feel body aches or discomfort in my body when caring for my child (for example, I normally feel pain when lifting or carrying the child).	1	1`	Retain & add an example
Physical	Overall	In general, I feel that caregiving is a physical strain, i.e. it requires much physical effort in performing the caregiving roles.	1	1`	Retain
Economic	Work	There have been adjustments in my income generation activities due to caregiving (for example, I had to turn	1	1`	Retain

		down a job, or I cannot engage in farming activities as much as I wish to).			
Economic	Expenses	There has been an increase in our family expenses due to the child's condition.	1	1`	Retain
Economic	Worry	I worry that I am unable to provide special facilities and services needed by my child, for example, providing for a special diet).	1	1`	Retain
Economic	Overall	In general, I feel that caregiving is a financial strain.	1	1`	Retain & Reword
Family	Relationships	Caring for my child often prevents me from attending to the needs of other family members (for example, most of my time is spent in providing care to the child with a disability).	1	1`	Combine with Q 14
Family	Relationships	There has been a disruption or upset of relationships within the family. (For example, other family members resent the attention I give to my child with a disability)	1	1`	Combine with Q 13
Family	Relationships	Parenting a child with a disability has caused more problems than I expected in my relationship with my spouse (significant other).	1	1`	Retain
Family	Overall	Overall, I feel that my family has been negatively affected by my child's condition.	1	1`	Retain
Child	Future	I worry about my child's future because of his/her condition.	1	1`	Combine with Q 18
Child	Future	I worry about whether my child will be able to live independently as an adult.	0.88	0.88	Combine with Q 17
Child	Behaviour	My child seems to cry more often than most children and this upsets me.	1	1	Retain
Child	Behaviour	I feel that my child asks for more help than he/she needs, and this upsets me.	1	1	Retain
Child	Behaviour	I often feel embarrassed by my child's behaviour.	0.88	0.88	Retain
Child	Stigma	I worry that my child often gets insulted/ridiculed by others (or other children).	1	1	Retain & reword
Child	Stigma	I am worried about bringing my child out to meet other people.	1	1	Combine with Q 24???
Child	Stigma	I worry that other people do not know how to treat my child.	1	1	Combine with Q 23
Child	Stigma	I often face embarrassing situations when travelling with the child.	1	1	Retain
Child	Communication	I worry that my child is not able to communicate as I would have expected.	1	1	Retain
Child	functioning	I feel sad that my child cannot do anything by him/herself like other children (for example, playing, toileting, going to school etc.).	0.88	0.88	Retain & reword
Child	Parental worry	I worry about something bad happening to my child when s/he is out of my care.	1	1	Retain
Child	Other	My child's sleeping and eating schedules were much harder to establish than I expected, and this worries me.	1	1	Retain
Child	Health	My child falls ill from time to time, and this worries me.	1	1	Retain
Child	Health	I worry that my child is not developing physically as other children of his/her age (for example, he/she is underweight).	1	1	Retain
Child	Health	I am worried that my child is not improving as much as I expected after receiving treatment/therapy.	0.88	0.88	Retain & reword

Social support	Family	I do get practical support from my family (for example, they help me with day-to-day activities such as bathing and dressing the child).	0.88	0.88	Retain
Social support	Family	I do get emotional support from my family (for example, I can discuss my child's problems with other family members).	1	1	Retain
Social support	Friends	My friends do help me with caring for the child (for example, I can always discuss with them some of the challenges I face in caring for my child).	1	1	Retain
Social support	Community	My neighbours do help me with caring for the child (for example, I can leave my child in their temporary care when I wish to go to the shops).	0.88	0.88	Retain & reword
Social support	Spouse	My spouse (significant other) helps me with the care of this child (for example, he/she supports me financially in caring for my child).	1	1	Retain
Social support	Overall	Overall, I feel that I have adequate help and support caring for my child.	1	1	Retain
Psychological	Anger	I feel angry when I think about the potential cause(s) of my child's condition.	0.88	0.88	Retain & reword
Psychological	Self-efficacy	I often feel uncertain about what to do about my child.	1	1	Redundant – similar to Q 42
Psychological	Self-efficacy	I feel that I should be doing more for my child.	1	1	Retain
Psychological	Self-efficacy	I feel confident in my ability to handle problems associated with caring for my child.	1	1	Redundant – similar to Q 40
Psychological	Guilt	I feel guilty about the potential cause(s) of my child's condition (for example, something I did, or my partner did during pregnancy caused my child's condition)	1	1	Retain
Psychological	Aetiology	I am focused on finding a specific reason for why this happened to my child.	0.75	0.72	Consider omission subject to testing.
Psychological	Aetiology	My child's condition could have been prevented.	0.625		Eliminate, similar to Q 39
Psychological	Aetiology	I worry that witchcraft caused my child's condition, or it is a sign of bad luck/omen.	0.75	0.72	Retain & reword
Psychological	Aetiology	I have come to terms with my child's condition.	0.88	0.88	Retain & reword
Psychological	Unappreciated	When I do things for my child, I get the feeling that my efforts are not well appreciated by my family.	0.88	0.88	Retain
Psychological	Hopelessness	Having caregiving responsibilities has created a feeling of hopelessness.	1	1	Retain
Social life	Confining	Because of caregiving, it is difficult to keep contact with relatives and friends (for example, I rarely get the opportunity to visit my relatives)	1	1	Redundant – similar to Q 51
Social life	Isolation	I feel isolated and alone (without friends) because of caring for the child.	1	1	Redundant – similar to Q 50
Social life	Time	I feel that I do not have enough time for my interests/hobbies because of the amount of time I spend caregiving.	1	1	Retain

Social life	Summative	Overall, I feel trapped by my caregiving responsibilities, i.e. my social life has negatively suffered because of providing care to my child.	1	1	Retain & reword
Overall	Overall	I feel completely overwhelmed by the caregiving role.	1	1	Retain
Overall	Overall	I have had more negative than positive experiences parenting a child with a disability.	0.88	0.88	Retain
Overall	Overall	I wish I could leave the care of the child to someone else.	0.88	0.88	Retain

The Median I-CV was 1.00 (IQR 0.88 –1.00). S-CVI/Ave and S-CVI-UA were .934 and .942 respectively. Version 3.0 thus consisted of 49 items phrased in the first person, i.e. self-report, and this was then subjected to cognitive debriefing.

5.6 Cognitive debriefing of Version 4.0

Before psychometric testing was done, the feasibility and acceptability of the ZCCS were explored through a cognitive debriefing exercise. A group of CCWCP thus completed the Version 3.0 questionnaire to examine the appropriateness, cultural acceptability, feasibility and relevance of the items certified by the panel of experts. It was also essential to translate the ZCCS into Shona as we intended to recruit caregivers of different educational status and English language proficiency. Two independent researchers performed the forward translations of the ZCCS from English to Shona. The co-supervisor then reconciled the forward translations into one version which was back-translated by another set of two independent translators. The PI reconciled the backward translations, and minor discrepancies were discussed with the co-supervisor to produce a Shona version of the ZCCS.

5.6.1 Sample

Ten caregivers were purposively recruited from both rural and urban areas. The sample size was based on recommendations from COSMIN guidelines on cognitive debriefing [150,532]. To ensure that different constituencies were included, the caregivers were of diverse education and economic background who were caring for children with various functional limitations (See Table 5.8 – Page 120).

Table 5.8: Demographics of cognitive debriefing sample

Variable	Attribute	Rural	Urban
Gender	Male	0	1
	Female	5	4
GMFCS Level of child under care	1	1	1
	2	1	1
	3	1	1
	4	1	1
	5	1	1
Relationship to child	Mother	2	3
	Father	0	1
	Aunt	1	0
	Grandmother	2	1
Level of education	None	2	0
	Primary	2	1
	Secondary	1	2
	Tertiary	0	1
Perceived financial adequacy	Very inadequate	1	2
	Inadequate	1	1
	Neutral	1	1
	Adequate	1	1
	Very adequate	1	0

N=10

5.6.2 Instrumentation

A self-designed questionnaire was used to collect the following caregivers' information: gender, relationship to the child, level of education and perceived level of income. The PI assessed the severity of children's disability using the gross motor function classification system (GMFCS). The GMFCS is a five-point ordinal measure for severity of CP with levels one and five being least and most severely affected individuals [484]. It is the most commonly used reliable and valid classification system [484,534]. Version 3.0 of the ZCCS was used for the cognitive debriefing exercise.

5.6.3 Procedure

Potential participants were identified by therapists working at the Children's Rehabilitation Unit (an urban tertiary hospital) and Mvurwi Rural District Hospital. Caregivers were interviewed as they presented for scheduled follow-up visits for their children. After explaining the aims of the study, consenting caregivers were required to sign a consent form or to give verbal consent in the presence of a witness. Caregivers completed the ZCCS in a quiet room; they rated the relevance and clarity of the items. Where appropriate, caregivers were especially encouraged to write down comments. After completion, the researcher interviewed the respondents to identify any problematic items and assess whether the ZCCS was a comprehensive and culturally-acceptable outcome measure. Caregivers' responses were documented manually on the completed form. The ZCCS was interviewer-administered for caregivers who were illiterate. We employed the teach-back method (TBM) technique to increase the validity of outcomes [535]. The researchers read out the questions and caregivers were requested to synthesise the meaning of the construct and to repeat the question in their own words before responding.

5.6.4 Data analysis

The frequency of responses to each question was calculated. Responses to the open-ended questions were recorded and were tabulated per item as the data were collected through structured interviews.

5.6.5 Ethical considerations

Caregivers were treated as autonomous agents as they were requested to consent before voluntarily participating. Interviews were held in a private room and data were de-identified through use of pseudo-names to preserve privacy and confidentiality. Caregivers were also provided with snacks after the interviews to compensate for their time. Further, pre-set criteria were utilized to elicit divergent views and to ensure that there were justice and equity across participants.

5.6.6 Results

Four items were omitted as these were deemed culturally inappropriate (n=3) and difficult to understand by caregivers (n=1). Further, two items which were conceptually overlapping were also merged into one item to improve clarity (Table 5.9- Page 122).

Table 5.9: Suggestions by caregivers

Item(s)	Recommendation
In general, I feel that my health has suffered because of the care I provide to the child.	Omit: the term "health" was considered by most caregivers [6/10] to be too generic as it has different colloquial meanings in the native language thus making the item confusing. Some caregivers [3/10] also found the item challenging to understand.
I feel that my child asks for more help than he/she needs, and this upsets me.	Omit – this item was deemed culturally inappropriate by most caregivers [9/10], thus the recommendation for the omission.
• I am worried about bringing my child out to meet other people. • I worry that other people do not know how to treat my child.	Most caregivers [7/10], felt that items were redundant as the underlying concept was the same. Action – adopted the item "I am worried about bringing my child out to meet other people" as it was clearer and culturally appropriate.
• I have had more negative than positive experiences parenting a child with a disability. • I wish if I could leave the care of the child to someone else.	Both items were omitted as they were deemed culturally inappropriate by most caregivers [7/10].
There have been adjustments in my income generation activities due to caregiving (for example, I had to turn down a job, or I cannot engage in farming activities as much as I wish to).	Need to use more simple language. Action – amended as follows: *"There have been changes in work plans due to caregiving (for example, "I had to turn down a job, or I cannot engage in farming activities as much as I used to").*

All caregivers were satisfied with the clarity of scoring and presentation of the items. One caregiver had this to say*, "I especially liked the pictures as they made it easy to understand the instructions"*. Younger caregivers [4/10] also suggested a bilingual presentation of the items to improve clarity. One caregiver expressed her difficulty in reading the text in the pure native language*, "… I find it difficult to read Shona fluently and would prefer if the questions were presented in both English and Shona…".* Based on feedback from caregivers, four items were dropped, and we changed illustrations for one of the items to reflect the experiences of caregivers residing in rural areas as well. These changes were implemented, and the Beta version of the ZCCS was finalised.

5.7 Discussion

We initially planned to extract candidate items from psychometrically sound tools identified through the SR as this is a recommended best practice in PROM development [39,40,536]. However, we applied a pragmatic approach as most of the retrieved tools [28/31] had limited evidence of content validity. Items with apparent high face and content validity were selected, and we utilised thematic and content analysis to group potential candidate items [537].

Content validity is defined as the extent to which items on an outcome measure adequately represent the construct and is considered the "most important" psychometric property [455,456]. Content validity is dependent on the socio-economic and cultural contexts, thus the need for qualitatively appraising the meaning of potential candidate items [39,40]. For example, the item on the FICD, "We had to postpone or cancel major holidays" is not applicable for caregivers residing in low resourced settings as caregivers grapple with a myriad of financial challenges associated with caring for a child with a disability [90,124]. To ensure a culturally and conceptually-robust outcome, we selected a panel of experts with diverse research and clinical practice expertise for content validation. Furthermore, we also critically appraised comments and suggestions by panellists as opposed to dropping/selecting an item entirely on I-CVI or modified Kappa indices as recommended by Polit et al. [529,538]. For instance, we combined conceptually equivalent items into one item irrespective of the high I-CVIs.

The scale content validity indices improved from S-CVI/Ave =.913 and S-CVI-UA= .912 to .934 and .942 respectively after the second round of panel of experts. The improvement illustrates the value of multiple assessments to improve content validity [39,40]. Addition of context-specific examples based on recommendations from the first round might have led to improved CVIs. For the second round, we selected panellists who had provided extensive input in the first round and a new set of panellists, and this could have led to the improved ICVIs and S-CVIs [531,533,538]. To increase the content validity, we also elicited views of caregivers of children with CP. We selected caregivers of different literacy and SES to assess the comprehensibility, relevance and clarity of the items and scoring instructions. Through this process, a further four items were excluded, and we also amended the presentation of the items. The importance of triangulation of input from target users, panel of experts & SR in developing a PRO can never be over-emphasised [39,40,501,539] and resulted in a credible alpha version of the ZCCS for further validation.

5.8 Conclusion

The robust content validation process resulted in a 45-item questionnaire which captured the range of experiences of caregivers of children with a disability in a low-resource setting. Item omission, addition and modification were guided by I-CVI indices and qualitative appraisal of comments and

suggestions by a panel of experts and target PRO users (caregivers). The 45-item version was then administered to a broad cross-section of caregivers to further evaluate psychometric properties such as structural validity, reliability, concurrent and construct validity (See Chapters Six & Eight).

6 Chapter 6: ZCCS structural validity and reliability evaluation

6.1 Introduction

The first stage in developing a new instrument is to establish content validity, and this was done in Chapter Five. The next stage was to test the structural validity and the reliability of this instrument. This chapter describes the further item reduction of the alpha version of the ZCCS based on factorial validity, and reliability testing through internal consistency and test-retest reliability evaluation.

6.2 Methods

6.2.1 Sampling

Primary caregivers who met the criteria of being unpaid for their caregiving role, living with a child who had been diagnosed with CP [540] and able to understand both written and spoken English and Shona were recruited. Caregivers were excluded if they had a confirmed psychiatric diagnosis according to the caregivers' medical records.

Although there are no clear guidelines for sampling estimates for CTT [155,169,179], some authors recommend an optimum ratio of 5-20 candidates per item for factor analysis [155,168,169,179,541]. To test the 45 items, it was anticipated that 400 participants would thus suffice. Additionally, we also set to recruit at least 50 participants to evaluate the stability (test-retest reliability) of the ZCCS [169,179].

6.2.2 Instrumentation

A self-designed demographic questionnaire was developed to elicit the following caregivers' information: age, educational level, employment status, place of residence (rural vs urban), gender and perceived SES. These personal factors were important in the evaluation of item and scale invariance during Rasch analysis.

The alpha version of the ZCCS is a 45-item outcome which was developed through a synthesis of findings from a systematic review, qualitative interviews and content validation process by a panel of experts. Caregiver burden is rated on a 5-point scale with responses ranging from strongly disagree=1 to strongly agree=5. Negatively worded items were reverse coded for analysis.

6.2.3 Procedure

6.2.3.1 Ethical considerations

Ethical approval for the study was granted by the University of Cape Town Ethical Review Board (Ref: 122/2016), Harare Central Hospital Ethics Committee (Ref: HCHEC 110316/18), City of Harare Health Services, and provincial medical directors of Mashonaland West and Central provinces and the

Medical Research Council of Zimbabwe (Ref: MRCZ/A/2072) (See Appendices 11.5, 11.6, 11.8, 11.9, 11.10, 11.10 and 11.11 respectively). Finally, written consent was sought from the caregivers before data collection.

6.2.3.2 Research team training

The PI recruited eight (8) research assistants who assisted with the recruitment of participants and administration of the ZCCS. To standardise the data collection procedures, the PI conducted a data collection familiarisation workshop. The study objectives were articulated, and thereafter, the PI explained the concepts enshrined in the ZCCS and elaborated on the scoring instructions. Research assistants were trained on how to approach prospective participants, obtaining participant consent (See Appendix 11.12 - Page 309) and administering the ZCCS through an interview should the need arise. The PI utilised didactic and role play teaching methodologies in training the research assistants. All research assistants were qualified Physiotherapy (6), and Occupational Therapy (2) interns with prior experience with data collection and all had previously completed individual dissertations and had assumed roles of research assistants in large-scale studies.

6.2.3.3 Data collection procedures

After obtaining either written or oral consent, the research team administered the ZCCS to participants. Questionnaires were self-administered, and the research team interview administered the ZCCS to caregivers who were illiterate. The research assistants read out the questions and filled in the responses on behalf of the participants. It was not always possible to get a private room to conduct the interviews, but the research team made all efforts to interview participants in seclusion. Respondents were also requested to indicate their willingness to complete the ZCCS after two weeks to evaluate the test-retest reliability. The research team drafted a list of participants who were interested in the re-test and contacted them through their mobile numbers a week before the re-test to confirm availability. The same data collection procedures as at baseline were followed in evaluating the ZCCS test-retest reliability.

6.2.4 Data analysis plan

Exploratory Factor Analysis (EFA) and Rasch analysis were used for the preliminary item bank reduction. Thereafter, the Generalized Least Squares Common Factor Analysis (FA) method was used for item reduction. The analysis was performed in the following sequence: item descriptive statistics; assessment of EFA testing assumptions; factor extraction; unrotated solutions; rotated solutions and finally the interpretation of factors.

6.2.5 Summary of structural validity testing plan

Outlined in Table 6.1 below and Table 6.2 (Page 128) is a summary of the EFA plan and the Rasch-based item reduction plan respectively. Items failing to meet the following criteria were subject to omission: ITC≤ 0.3, and item residuals ≥ 2.5.

Table 6.1: ZCCCS factor analysis plan

Stage	Hypothesis/notes	Evaluative criteria
Testing of assumptions	Normality – data should be normally distributed	A non-significant Shapiro Wilkson Test statistic (p>0.05)
	Adequate inter-item correlations & absence of multicollinearity	i. Adequate inter-item correlations, i.e. r≥0.3 ii. items with excessively high inter-item correlations, i.e. r≥0.9 should be removed from the analysis iii. Item-total correlation; ITC≥.4
	Adequate sample variation	i. KMO≥ 0.5. [KMO values are interpreted as .5-.7- mediocre, .7-.8- good, .8-.9- great & >.9 – superb] ii. Participants to item ratio of at least 10:1
	Missing data	i. Missing values should occur in a non-random manner ii. Omit missing values to prevent over-estimation
	Factorability of the data	i. A significant Bartlett test of Sphericity- p≤ 0.05 means that the correlation matrices statistically different from an identity matrix, i.e. there is a correlation between variables within the factor
Factor extraction method	Various factor extraction methods can be applied, and they can yield different results	ii. Test several methods to identify a parsimonious solution iii. Model selection is an iterative process and should be guided by theory
Dimensionality /factor retention	Use multiple methods for factor retention	i. Kaiser's Criterion- retain factors with eigenvalues ≥ 1 ii. Horn's Parallel method- Create a set of random variables, and a factor is retained if the magnitude of the random eigenvalue is less that of Kaiser eigenvalue iii. Cartel's Scree plot- inspect for breaks in continuity on the eigenvalues vs a number of factors plot. Retain all factors just before the point of inflexion
Factor rotation	Apply oblique rotation	i. Compared to Promax, direct Oblimin rotation does not allow high correlation between factors ii. Apply both methods and select the parsimonious solution
Factor interpretation	Qualitatively analyse the conceptual meaning of items loading onto a factor	i. A meaningful factor should contain at least three (3) items ii. Factor naming is an "art"

Results for both CTT and Rasch indices were also qualitatively appraised before deleting an item to ensure the preservation of content validity. Items meeting both CTT and Rasch requirements then underwent factor analysis to evaluate the structural validity of the ZCCS further.

Table 6.2: ZCCS Rasch-based item reduction plan

Scale trait	Method of analysis	Hypothesis/Evaluative criteria
Response distribution	Frequency plot of actual responses across all items and response options	Check that each possible category of item/response is endorsed; ideal to have at least 5 cases in each
Person fit	Chi-square, fit residual transformed to a standardised (Z) score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all items scored by a person
Item fit	Chi-square, fit residual transformed to a Z-score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all persons for a given item
Item trait interaction	Chi-square probability	p>0.05 tests whether the items are working as expected across the class intervals for the trait (the hierarchical ordering of items)
Individual person fit	Chi-square analysis	Values should fall within ±2.5 if the data fit the Rasch model

Adapted from the following sources: [187-195,207]

6.3 Results

6.3.1 Demographic information of the respondents and their children

As seen in Table 6.3 (Page 129), most children were males (60.5%), resided in urban areas (87.2%) with a median age of 4.1 years (interquartile range: 4). There was an equal distribution of children within different disability classes. Most caregivers were mothers to the children (89.2%), educated (98.5%), unemployed (80.3%), and had provided care for at least 48 months.

Table 6.3: Characteristics of the study participants

Variable	Attribute	Children with CP n=461	Caregivers, n=461
Gender	Male	279 (60.5)	452 (98.0)
	Female	182 (39.5)	9 (2.0)
Age in years	*Median [Q_1-Q_3]	4.1 [2.2-6.2]	34.8 (SD 10.6)
Place of residence	Urban	402 (87.2)	402 (87.2)
	Rural	59 (12.8)	59 (12.8)
GMFCS level	1	99 (21.6)	
	2	55 (12)	
	3	93 (20.3)	
	4	91 (19.9)	
	5	120 (26.2)	
Relationship to child	Mother		411 (89.2)
	Grandmother		29 (6.3)
	Aunt		12 (2.6)
	Father/grandfather		9 (2.0)
Duration of caregiving	Median [Q_1-Q_3]		48 [25-80]
Marital status	Married		353 (76.6)
	Divorced		51 (11.1)
	Widower/widow		35 (7.6)
	Never lived together		11 (2.4)
	Single		9 (2.0)
Education	Secondary		357 (77.4)
	Primary		83 (18)
	Tertiary		10 (2.2)
	None		7 (1.5)
Employment	Unemployed		370 (80.3)
	Informally employed		47 (10.2)
	Formally employed		25 (5.4)
	Farming		19 (4.1)

Perceived financial status	Very inadequate		177 (38.4)
	Inadequate		73 (15.8)
	Neutral		177 (38.4)
	Very adequate		20 (4.3)
	Adequate		14 (3)

6.3.2 Item descriptive statistics

As seen in Table 6.4 below, there were few missing responses and items 24 and 22 had the most extreme scores.

Table 6.4: ZCCS item descriptives

Item	N	Mean	SD	Missing		No. of Extremes	
				n	%	Low	High
1PHtimeampneeds	458	2.7	1.3	3	.7	0	0
2PHfuture	460	3.1	1.4	1	.2	0	0
3PHsleep	460	2.8	1.4	1	.2	0	0
4PHtired	460	2.7	1.3	1	.2	0	0
5PHpain	460	3.1	1.4	1	.2	0	0
6PHphystrain	461	3.3	1.5	0	.0	0	1
7ECworkadj	461	3.7	1.3	0	.0	0	0
8ECfamexp	461	3.7	1.2	0	.0	0	0
9ECspefaci	459	3.8	1.2	2	.4	0	0
10ECfinstrain0	460	3.7	2.3	1	.2	0	1
11FAMrelationships	461	3.1	1.4	0	.0	0	0
12FAMspouse	458	3.1	1.4	3	.7	0	0
13FAMoverall	460	2.9	1.3	1	.2	0	0
14CHIfuture	461	4.2	1.1	0	.0	50	0
15CHIcry	459	3.0	1.4	2	.4	0	0
16CHIbehaviour	461	2.7	1.3	0	.0	0	0
17CHIinsulted	460	3.4	1.4	1	.2	0	0

18CHIstigma	460	3.4	1.4	1	.2	0	0
19CHIembrassing	459	3.0	1.4	2	.4	0	0
20CHIcommunication	458	3.6	1.4	3	.7	0	0
21CHIcannotdo	460	3.9	1.3	1	.2	0	0
22CHIsomethingbad	461	4.0	1.1	0	.0	65	0
23CHIsleep	461	3.1	1.4	0	.0	0	0
24CHIill	461	3.2	1.4	0	.0	0	0
25CHdevelopment	460	3.5	1.3	1	.2	0	0
26CHIimproving	459	2.9	1.3	2	.4	0	0
27SSpracfam	459	3.2	1.4	2	.4	0	0
28SSemotfam	460	2.7	1.3	1	.2	0	0
29SSfrehelp	458	3.0	1.3	3	.7	0	0
30SSneig	458	3.4	1.4	3	.7	0	0
31SSspouse	458	2.5	1.3	3	.7	0	0
32SSoveral	456	3.1	1.2	5	1.1	0	0
33PSYangry	459	3.5	1.4	2	.4	0	0
34PSYconfidence	456	2.1	1.1	5	1.1	0	66
35PHYguilt	461	2.8	1.3	0	.0	0	0
36PSYwitchcraft	460	3.1	1.4	1	.2	0	0
37PSYacceptance	457	2.2	1.2	4	.9	0	0
38PSYunappreciated	461	2.9	2.3	0	.0	0	1
39PSYhopelessness	461	2.8	1.3	0	.0	0	0
40PSYaccomodation	460	3.0	1.5	1	.2	0	0
41PSYspace	461	3.3	1.4	0	.0	0	0
42SLcontact	461	3.2	1.4	0	.0	0	0
43SLhobbies	461	3.1	1.3	0	.0	0	0
44SLoverall	461	2.9	1.3	0	.0	0	0
45SUMoverwhelmed	459	3.3	1.3	2	.4	0	0

6.3.3 Stage 1 – testing for assumptions

As demonstrated in Table 6.5 below, data were normally distributed, exhibited adequate sampling adequacy (KMO =.903), and the correlation matrix was factorable (Bartlett Test of Sphericity was statistically significant; <0.001) thus validating the suitability of EFA. Additionally, the sample was large (N=461), there were low missing and extreme values (See Table 6.4), and only nine (9) items had ITC< 0.3 (See Table 6.6).

Table 6.5: EFA assumptions- ZCCS evaluation

Attribute	Statistic	Parameters
Normality	Shapiro- Wilk test	W= 0.987, df=451, p=0.10
Sampling adequacy	The Kaiser-Meyer-Olkin Measure of Sampling Adequacy	.903
Matrix identity	Bartlett Test of Sphericity	χ^2 (df 990) = 7407.9, p<0.001

6.3.4 Items correlation matrix

Items 27, 28, 29, 30, 31, 34, 35, 37 and 38 (highlighted) had item-total correlation of less than .3 and were thus candidates for removal.

Table 6.6: ZCCS item -total correlations

Item	ITC	Item	ITC	Item	ITC
1PHtime& needs	0.527	16CHIbehaviour	0.401	31SSspouse	0.250
2PHfuture	0.498	17CHIinsulted	0.437	32SSoveral	0.324
3PHsleep	0.544	18CHIstigma	0.541	33PSYangry	0.349
4PHtired	0.581	19CHIembrassing	0.528	34PSYconfidence	0.041
5PHpain	0.531	20CHIcommunication	0.466	35PHYguilt	0.279
6 PHphystrain	0.489	21CHIcannot do	0.558	36PSYwitchcraft	0.319
7ECworkadj	0.585	22CHIsomethingbad	0.463	37PSYacceptance	0.013
8ECfamexp	0.591	23CHIsleep	0.526	38PSYunappreciated	0.273
9ECspefaci	0.605	24CHIill	0.520	39PSYhopelessness	0.533
10ECfinstrain	0.434	25CHdevelopment	0.374	40PSYaccomodation	0.527
11FAMrelationships	0.604	26CHIimproving	0.500	41PSYspace	0.485
12FAMspouse	0.499	27SSpracfam	0.176	42SLcontact	0.632
13FAMoverall	0.570	28SSemotfam	0.233	43SLhobbies	0.628
14CHIfuture	0.411	29SSfrehelp	0.219	44SLoverall	0.642
15CHIcry	0.488	30SSneig	0.217	45SUMoverwhelmed	0.616

6.3.5 Item fit assessment through Rasch analysis

Item fit was assessed by evaluating the magnitude of residuals and global fit with the PCM. Item fit is the extent to which a specific item satisfies the requirements of the Rasch model, i.e. the extent to which it is reflective of the domain under investigation [187-189,192,195]. In the present study, items with a perfect fit indeed measure caregiver burden. Items with residuals ≥ 2.5 and statistically significant deviations after Bonferroni corrections (depicted under F-stat/p column) were flagged, and items 7, 9-11, 13, 27- 31, 33-35, 37, 42-45, were candidates for possible elimination (Table 6.7.

Table 6.7: ZCCS item fit statistics

item description	Location	SE	Fit Resid *	Chi Sq	DF 2	Chi sq p	F-stat	DF-1**	DF-2**	F stat p	Fit
1PHtime&needs	0.355	0.042	-1.05	5.0	6	0.539	0.907	6	424	0.490	fit
2PHfuture	0.011	0.04	-0.31	10.4	6	0.110	1.839	6	424	0.090	fit
3PHsleep	0.229	0.04	-0.22	12.8	6	0.047	2.012	6	424	0.063	fit
4PHtired	0.282	0.042	-1.89	17.5	6	0.008	3.511	6	424	0.002	fit
5PHpain	0.056	0.04	-0.77	4.8	6	0.573	0.972	6	424	0.444	fit
6 PHphystrain	-0.114	0.043	-1.70	15.1	6	0.019	3.301	6	424	0.003	fit
7ECworkadj	-0.374	0.043	-2.98	23.4	6	0.001	5.251	6	424	0.000	misfit
8ECfamexp	-0.524	0.044	-2.44	16.1	6	0.013	3.677	6	424	0.001	fit
9ECspefaci	-0.529	0.045	-2.80	18.7	6	0.005	4.24	6	424	0.000	misfit
10ECfinstrain0	-0.473	0.045	-3.21	32.1	6	0.000	7.682	6	424	0.000	misfit
11FAMrelationships	0.052	0.041	-3.62	24.5	6	0.000	6	6	424	0.000	misfit
12FAMspouse	0.058	0.039	-1.15	7.7	6	0.257	1.558	6	424	0.158	fit
13FAMoverall	0.192	0.042	-2.71	19.7	6	0.003	4.51	6	424	0.000	misfit
14CHIfuture	-0.666	0.05	-0.27	6.4	6	0.379	1.045	6	424	0.396	fit
15CHIcry	0.082	0.038	0.10	3.3	6	0.776	0.526	6	424	0.788	fit
16CHIbehaviour	0.308	0.041	0.71	13.1	6	0.042	2.393	6	424	0.028	fit
17CHIinsulted	-0.141	0.039	1.09	3.8	6	0.709	0.502	6	424	0.807	fit
18CHIstigma	-0.181	0.04	-1.91	11.5	6	0.074	2.416	6	424	0.026	fit

19CHIembrassing	0.046	0.04	-1.41	9.4	6	0.152	2.05	6	424	0.058	fit
20CHIcommunication	-0.182	0.039	0.33	1.9	6	0.926	0.284	6	424	0.945	fit
21CHIcannot do	-0.575	0.044	-1.96	18.7	6	0.005	3.752	6	424	0.001	fit
22CHIsomething bad	-0.563	0.048	-0.66	5.3	6	0.507	0.829	6	424	0.548	fit
23CHIsleep	0.045	0.039	0.34	6.0	6	0.422	0.995	6	424	0.428	fit
24CHIill	-0.086	0.041	0.05	4.1	6	0.660	0.69	6	424	0.658	fit
25CHdevelopment	-0.194	0.041	1.89	7.4	6	0.286	1.161	6	424	0.326	fit
26CHIimproving	0.136	0.04	-0.64	10.1	6	0.123	1.863	6	424	0.086	fit
27SSpracfam	0.064	0.039	7.07	61.5	6	0.000	8.753	6	424	0.000	misfit
28SSemotfam	0.355	0.041	4.72	31.6	6	0.000	4.505	6	424	0.000	misfit
29SSfrehelp	0.099	0.041	5.65	45.8	6	0.000	6.476	6	424	0.000	misfit
30SSneig	-0.153	0.04	5.42	37.9	6	0.000	5.697	6	424	0.000	misfit
31SSspouse	0.385	0.039	3.33	45.6	6	0.000	6.777	6	424	0.000	misfit
32SSoveral	0.035	0.045	1.71	19.6	6	0.003	3.077	6	424	0.006	fit
33PSYangry	-0.135	0.039	2.69	11.2	6	0.081	1.9	6	424	0.079	fit
34PSYconfidence	0.79	0.046	4.96	95.2	6	0.000	13.323	6	424	0.000	misfit
35PHYguilt	0.331	0.041	4.15	26.0	6	0.000	3.7	6	424	0.001	misfit
36PSYwitchcraft	0.056	0.038	4.58	25.5	6	0.000	3.739	6	424	0.001	misfit
37PSYacceptance	0.511	0.04	8.03	117.9	6	0.000	15.355	6	424	0.000	misfit
38PSYunappreciated	0.231	0.04	2.65	16.6	6	0.011	2.482	6	424	0.023	fit
39PSYhopelessness	0.213	0.043	-1.62	15.9	6	0.014	3.258	6	424	0.004	fit
40PSYaccomodation	0.079	0.036	-0.19	4.4	6	0.621	0.767	6	424	0.596	fit
41PSYspace	-0.125	0.038	1.06	5.9	6	0.439	0.973	6	424	0.443	fit
42SLcontact	0.026	0.039	-3.40	27.5	6	0.000	6.479	6	424	0.000	misfit
43SLhobbies	0.046	0.041	-3.15	28.1	6	0.000	6.635	6	424	0.000	misfit
44SLoverall	0.094	0.042	-3.74	41.0	6	0.000	10.427	6	424	0.000	misfit
45SUMoverwhelmed	-0.154	0.042	-2.42	22.5	6	0.001	4.698	6	422	0.000	misfit

**Fit residuals ** DF=Degrees of freedom*

6.3.6 Item exclusion based on EFA and Rasch analysis

After synthesis of the results for item-total correlation (ITC) and item fit based on Rasch analysis, we excluded the following items: 27-30. We then performed EFA on the remaining item set and misfitting items were deleted stepwise as per the analysis plan as previously described (Table 6.8).

Table 6.8 : Results of item exclusion on item correlations and item fit statistics

Item description	FitRes *	Discrimination	ITC	Decision	Item description	FitRes *	Discrimination	ITC	Decision
1PHtime&needs	-1.05	optimal	0.53	retain	24CHIill	0.05	optimal	0.52	retain
2PHfuture	-0.31	optimal	0.50	retain	**25CHdevelopment**	1.89	optimal	0.37	retain
3PHsleep	-0.22	optimal	0.54	retain	**26CHIimproving**	-0.64	optimal	0.5	retain
4PHtired	-1.89	optimal	0.58	retain	**27SSpracfam**	7.07	under	**0.18**	**omit**
5PHpain	-0.77	optimal	0.53	retain	**28SSemotfam**	4.72	under	**0.23**	**omit**
6 PHphystrain	-1.70	optimal	0.49	retain	**29SSfrehelp**	5.65	under	**0.22**	**omit**
7ECworkadj	-2.98	marginal over	0.59	retain	**30SSneig**	5.42	under	**0.22**	**omit**
8ECfamexp	-2.44	optimal	0.59	retain	**31SSspouse**	3.33	under	0.25	retain
9ECspefaci	-2.80	marginal over	0.61	retain	**32SSoveral**	1.71	optimal	0.32	retain
10ECfinstrain0	-3.21	over	0.43	retain	**33PSYangry**	2.69	marginal under	0.35	retain
11FAMrelationships	-3.62	over	0.60	retain	**34PSYconfidence**	4.96	under	**0.04**	**omit**
12FAMspouse	-1.15	optimal	0.50	retain	**35PHYguilt**	4.15	under	**0.28**	**omit**
13FAMoverall	-2.71	marginal over	0.57	retain	**36PSYwitchcraft**	4.58	under	**0.32**	**omit**
14CHIfuture	-0.27	optimal	0.41	retain	**37PSYacceptance**	8.03	under	**0.01**	**omit**
15CHIcry	0.10	optimal	0.49	retain	**38PSYunappreciated**	2.65	marginal under	0.27	retain
16CHIbehaviour	0.71	optimal	0.40	retain	**39PSYhopelessness**	-1.62	optimal	0.53	retain

17CHIinsulted	1.09	optimal	0.44	retain	**40PSYaccomodation**	-0.19	optimal	0.53	retain
18CHIstigma	-1.91	optimal	0.54	retain	**41PSYspace**	1.06	optimal	0.49	retain
19CHIembrassing	-1.41	optimal	0.53	retain	**42SLcontact**	-3.40	over	0.63	retain
20CHIcommunication	0.33	optimal	0.47	retain	**43SLhobbies**	-3.15	over	0.63	retain
21CHIcannotdo	-1.96	optimal	0.56	retain	**44SLoverall**	-3.74	over	0.64	retain
22CHIsomethingbad	-0.66	optimal	0.46	retain	**45SUMoverwhelmed**	-2.42	optimal	0.62	retain
23CHIsleep	0.34	optimal	0.53	retain					

*Fit Residual ** Item-total correlation

6.3.7 Stage 2 – factor extraction

Exploratory factor analysis was then performed on the remaining 37 items. Kaiser criterion (Table 6.9) and Monte Carlo parallel (Table 6.10) extraction methods supported the retention of four (4) factors whereas the output from the Scree plot was indeterminate (Figure 6-1).

Table 6.9: Initial eigenvalues -ZCCS

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	10.854	30.149	30.149	4.870	13.526	13.526
2	1.801	5.002	35.151	6.316	17.545	31.071
3	1.579	4.386	39.537	1.182	3.284	34.355
4	1.530	4.249	43.786	1.325	3.682	38.037
5	1.343	3.731	47.517	1.075	2.987	41.024
6	1.268	3.522	51.040	.945	2.624	43.648
7	1.119	3.109	54.148	.869	2.415	46.063
8	1.061	2.947	57.096	.690	1.916	47.979

Table 6.10: Random Eigen values - ZCCS

Eigenvalue #	Kaiser criterion value	*Random Eigenvalue (SD)	Decision
1	10.854	1.57 (SD.037)	Retain
2	1.801	1.499 (SD.029)	Retain
3	1.579	1.447 (SD .026)	Retain
4	1.530	1.403 (SD .022)	Retain
5	1.343	1.362 (SD.021)	Reject
6	1.268	1.326 (SD.019)	Reject
7	1.119	1.292 (SD.019)	Reject
8	1.061	1.260 (SD .018)	Reject

*Values for random eigenvalues are based on the following parameters: 36 variables, 461 participants and 1000 replications. A factor is retained if the magnitude of the random eigenvalue is less than that of Kaiser eigenvalue [155,169].

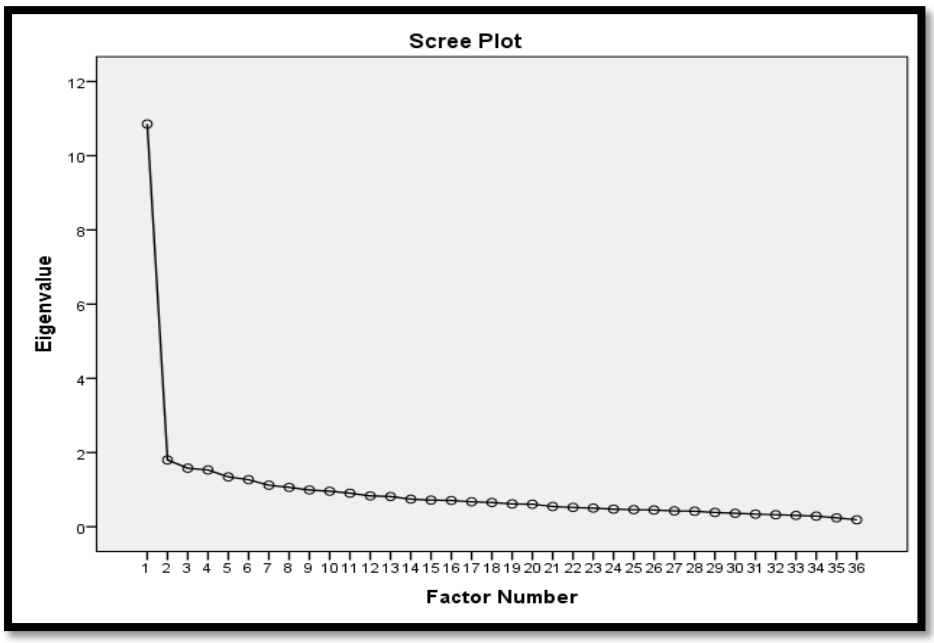


Figure 6-1: Scree plot – ZCCS

6.3.8 Stage 3 – factor rotation and interpretation

Item reduction using the generalised least square methods yielded a 33-items, four-factor solution confirming the multidimensionality of caregiver burden (Table 6.11- Page 138).

Table 6.11: ZCCS pattern matrix

Item	1. Physical & economic burden	2. Concerns for the child	3. Family relations	4. Community participation
10ECfinstrain0	.819			
8ECfamexp	.785			
5PHpain	.721			
9ECspefaci	.645			
4PHtired	.628			
7ECworkadj	.548			
6 PHphystrain	.506			
2PHfuture	.411			
45SUMoverwhelmed	.337			
1PHtime&amp	.330			
17CHIinsulted		.608		
18CHIstigma		.586		
23CHIsleep		.553		
25CHdevelopment		.529		
14CHIfuture		.522		
21CHIcannot do		.521		
20CHIcommunication		.512		
15CHIcry		.505		
22CHIsomethingbad		.414		
24CHIill		.398		
36PSYwitchcraft		.364		
3PHsleep		.346		
13FAMoverall			.819	
12FAMspouse			.684	
11FAMrelationships			.556	
16CHIbehaviour			.412	
31SSspouse			.403	
35PHYguilt			.370	
19CHIembrassing			.334	
43SLhobbies				.956
44SLoverall				.890
42SLcontact				.623

N=461

Factors retained include physical & economic burden (10 items), concerns for the child (12 items), family relations (7 items) and community participation (4 items) which were retained upon inspection of structure and pattern matrices (Table 6.11 and Table 6.12 respectively) and discussion with the supervisors. Eighteen items exhibited cross-loadings > 0.4 as seen in the structure matrix. Factors underwent Promax (oblique) rotation method to improve interpretability. Furthermore, most items [24/33] had high factor loadings > 0.4 [range: 0.405 – 0.956].

Table 6.12: ZCCS structure matrix:

Item	1. Physical & economic burden	2. Concerns for the child	3. Family relations	4. Community participation
10ECfinstrain0	.783	.510		.423
8ECfamexp	.727			.412
9ECspefaci	.691	.567		
5PHpain	.661			
4PHtired	.660		.437	.434
7ECworkadj	.660	.463		.494
6 PHphystrain	.616		.467	.485
45SUMoverwhelmed	.577	.464	.445	.494
2PHfuture	.532		.445	.412
1PHtime&amp	.497			.407
21CHIcannot do	.533	.647		.417
18CHIstigma	.414	.642		.406
23CHIsleep		.569		
20CHIcommunication		.560		
14CHIfuture		.552		
15CHIcry		.547		
22CHIsomethingbad	.470	.546		
17CHIinsulted		.545		
25CHdevelopment		.531		
3PHsleep	.508	.524		
24CHIill	.439	.518		
36PSYwitchcraft		.364		
13FAMoverall	.502		.839	.404
11FAMrelationships	.528	.432	.713	.527
12FAMspouse			.699	
19CHIembrassing	.428	.465	.493	

16CHIbehaviour			.484	
31SSspouse			.370	
35PHYguilt			.3334	
43SLhobbies	.521	.435		.899
44SLoverall	.531	.435	.440	.880
42SLcontact	.487	.453	.424	.714
40PSYaccomodation				.478
Extraction Method- Generalized Least Squares. Rotation Method- Promax with Kaiser Normalization.				

N=461

6.3.9 ZCCS Internal consistency

The ZCCS yielded excellent internal consistency (IC) ratings at both sub-scale (α range: .765-.841) and scale level (α=.925). Deletion of any of the items did not produce improvements in the scale IC thus suggesting the reliability of items (See Table 6.13 and Table 6.14).

Table 6.13: ZCCS Item-total statistics

Item	Scale Mean if Item Deleted	Scale Variance if Item Deleted	Corrected Item-Total Correlation	Cronbach's Alpha if Item Deleted
1PHtime&amp	104.0	534.7	.491	.923
2PHfuture	103.6	532.7	.499	.922
3PHsleep	103.9	531.4	.522	.922
4PHtired	104.0	530.3	.567	.922
5PHpain	103.6	531.1	.528	.922
6 PHphystrain	103.4	531.1	.571	.922
7ECworkadj	103.0	529.3	.598	.921
8ECfamexp	103.0	531.5	.578	.922
9ECspefaci	102.8	532.4	.583	.922
10ECfinstrain0	103.1	529.0	.632	.921
11FAMrelationships	103.6	525.5	.625	.921
12FAMspouse	103.6	531.8	.504	.922
13FAMoverall	103.8	529.2	.583	.921
14CHIfuture	102.5	544.4	.420	.923
15CHIcry	103.7	533.1	.467	.923
16CHIbehaviour	104.0	539.7	.412	.923
17CHIinsulted	103.2	537.9	.405	.924

18CHIstigma	103.3	530.3	.545	.922
19CHIembrassing	103.7	530.6	.529	.922
20CHIcommunication	103.0	533.8	.472	.923
21CHIcannot do	102.8	532.7	.560	.922
22CHIsomethingbad	102.6	539.9	.483	.923
23CHIsleep	103.6	532.7	.485	.923
24CHIill	103.5	535.1	.470	.923
25CHdevelopment	103.2	541.0	.385	.924
31SSspouse	104.2	551.8	.202	.926
35PHYguilt	103.9	547.1	.280	.925
36PSYwitchcraft	103.6	544.1	.304	.925
42SLcontact	103.5	526.0	.588	.921
43SLhobbies	103.6	527.1	.604	.921
44SLoverall	103.7	526.0	.630	.921
45SUMoverwhelmed	103.4	528.7	.598	.921
40PSYaccomodation	103.7	531.4	.466	.923

N=461

Table 6.14: ZCCS Subscales internal consistency values

	Physical & economic burden	**Concerns for the child**	**Family relations**	**Community participation**	**Scale Level**
α	.841	.828	.765	.813	.925
ICC (95% CI)	.841 (.765: .900)	.828 (.804: .851)	.765 (.730: 796)	.813 (.784: .840)	.925 (.914: .934)

N=461

6.3.10 ZCCS test-retest reliability

The ICC (95% CI) for summed ZCCS scores at baseline and after four weeks was .880 (0.793: 0.930). Data were analysed for 54 participants.

6.4 Discussion

6.4.1 Structural validity

We believe that the 33-items solution has robust evidence for structural validity. Firstly, the item pool was a product of findings from a systematic review, qualitative interviews and input from a panel of experts. The exclusion of only 12 items from the original pool of 45 items is further testimony of the

robust content validation process. Secondly, data adequately met the pre-requisites for EFA, i.e. it was normally distributed, had maximal variation (KMO=0.903) and the Bartlett Test of Sphericity was non-significant [165,171]. Thirdly, the use of multiple factor extraction methods also increased the confidence in the internal study validity. Although Kaisers' criterion of eigenvalues is the most popular factor extraction method [164,165], it often overstates the number of factors extracted, and there has been advocacy for usage of multiple methods for factor extraction [166,169, 175]. However, use of the Kaiser methodology resulted in 57% variation explained by the eight factors extracted as opposed to 48% variance explained by the four factors retained through the amalgamated use of both the Kaiser and Horn parallel analysis methods. Nevertheless, in addition to attaining maximal variation, it is also important to attain a parsimonious solution in EFA [155,165,171,175].

Fourthly, the use of both EFA and Rasch analysis for preliminary item selection increased the robustness of the item reduction process. Given the philosophical and methodological differences between CCT and IRT item reduction techniques, there is a growing impetus in utilising both methodologies to increase psychometric rigour [191-197]. Finally, qualitative analysis and stepwise deletion of the item with low factor loadings also ensured the preservation of the content validation of the ZCCS. For example, item 8 (There has been an increase in our family expenses due to the child's condition) loaded poorly onto the economic and physical burden factor. However, the item was retained for subsequent analysis as the item was deemed conceptually essential. After stepwise deletion of misfitting items, the item factor loading increased substantially (r=.785). This is an example of the importance of analysing an item before removal based on factor loadings, and this also displays the inherent weakness of CTT methods, unlike Rasch modelling where items can be omitted based on multiple pre-set criterion [166,191,192].

6.4.2 Discussion of the four factors identified

The naming of putative factors is described as an "art" given the need to maintain simplicity and preservation of the conceptual meaning of items within the same factor [165,166,171]. The findings are consistent with previous studies [83,493-495] and the results of the earlier qualitative study (Chapter Four) which confirmed that caregivers were affected emotionally, physically, socially and financially. In addition, the four factors fit into the ICF framework in that child, and caregiver well-being includes items mostly related to impairments such as pain or anxiety, and the family relationships and community participation factors reflect the participation restriction and environmental components. The economic and physical health items are not solely related to the environment but do pick up on the impact that environmental factors have on physical functioning.

6.4.2.1 Factor 1: Economic and physical health impact

The loading of items measuring financial burden (e.g. item 8 - …increase in family expenses…) and physical burden (e.g. item 4- I feel tired and exhausted because of caring for the child) onto the same factor exemplifies the complexity of naming factors [165,166,171]. Nevertheless, items in the financial domain had the highest loadings [range: .660 - .813], which demonstrates the impact of finances on caregivers' well-being. This is unsurprising given the current socio-economic turmoil in Zimbabwe, for example, 80% of the population survives on less than US$2 /day [505-507]. The high rate of unemployment and lack of social grants further exacerbate the financial burden as caring for a child with CP is associated with increased medical expenditure and usage of specialist services [23,492,493,505,506]. Furthermore, retention of all the original four items further testifies to the impact of the financial burden. The loading of the global burden rating item (item 45- Overall, I feel completely overwhelmed by the caregiving role) on to this factor is suggestive that financial strain is a pivotal predictor to caregiver burden [23,493]. Additionally, caregivers reported that caring for a child with a disability had affected their future, e.g. plans for having other children (Item 2). This can also be linked to the financial burden, which similarly emerged from the interviews (Chapter Four).

Caregivers also reported physical burden in this factor as they endorsed items on pain (item 5), fatigue (item 4), time pressure (item 1) and global physical burden rating (item 6). Most children with CP require assistance in ADLs, lifting and transfers, and this leads to the high burden of musculoskeletal disorders such as low back pain and joint pain in caregivers [89,499,542-544]. Further, the lack of mobility aids and appliances (due to high costs) predisposes caregivers to musculoskeletal disorders [493,494,498,544]. As reported elsewhere [18,83,124], most caregivers in the present study carried their children on their backs as they could not afford wheelchairs, and this predisposed them to musculoskeletal disorders.

6.4.2.2 Factor 2: Concerns for the child

It has been suggested that the HRQoL of the caregiver and the young child are interrelated and that the well-being of the one influences the other [545]. The items that load onto this factor indicate that this relationship is evident in children with CP as well. Diagnosis of CP is envisaged as a "traumatic experience" with some caregivers failing to adequately adjust to the excessive demands of the caregiving role [495,498,502]. Evidence from several systematic reviews has shown that caregivers are likely to suffer from poorer mental health as compared to caregivers of typically developing children (TDC) [16,218,543,546,547]. Caring for a TDC is challenging, and the dynamics become even more complicated when caring for a child with a life-long physical disability [124,471,548]. For instance, caregivers reported being worried about the future of the child, and the rate of physical development

and this was congruent with the earlier qualitative study. Further, child behavioural problems, excessive crying and communication problems have also been previously cited as critical predictors of caregiver burden [124,548]. Caregivers in the present study cited stigma as a source of distress. It is unfortunate that myths surrounding the causative agents of CP are still prevalent in Africa. As discussed previously (Section 4.7.3), the belief that witchcraft causes CP and bad luck are still common [17-23,493,498,503,504]. Some caregivers appear to share a similar belief by endorsing item 36 (I worry that my child's condition was caused by witchcraft or is a sign of evil luck/omen).

6.4.2.3 Factor 3: Family relationships

As reported elsewhere (Chapters Two and Four), caregivers experience alterations in family dynamics related to caring for a child with CP [285,308,550]. This accords with the Double ABC-X model which postulates that caregiver burden is a product of the continuous accumulation of stressors, which negatively affect family resources/ability to deal with distress and family perception of the caregiving situation [107-109]. Caregiving stress may lead to frequent arguments and in extreme cases to divorce [23,493,498,503]. Marriage breakdown associated with myths such as CP being caused by maternal promiscuity and being hereditary has also been reported in other low-resource settings [17-23,493,498,503,504]. Alterations in family relations may also be interlinked with the financial burden. For example, most resources are likely to be channelled towards meeting the needs of the child with CP, and this may, unfortunately, lead to tension with other family members [280,285,308,550,551]. The presence of child behaviour problems was also cited as a source of caregiver distress. Behaviour problems such as inappropriate urination, excessive crying, among others, have been mentioned as sources of caregiver burden [124,258,280,317,548,552]. Interestingly, child behaviour problem items cross-loaded on both the family relations and concerns for the child, and this further testifies to the impact of behavioural problems on caregiver burden [317].

6.4.2.4 Factor 3: Community participation

Caregiving is often associated with time pressure and a decrease in social networks [498]. Caregivers reported that caregiving had negatively affected their social life. Both enacted and societal stigma leads to decreased social networks and social participation thus adversely affecting caregivers' social life [7,85,87-89,498]. Further, financial burden and lack of respite care as support resources are also likely to diminish further caregivers' opportunities for pursuing hobbies [88,89,124]. Item 40 (challenges in finding accommodation) also loaded onto the social life domain. Although conceptually tangent to the rest of the items within the factor, the item unearths some of the challenges encountered by Zimbabwean caregivers. It is unfortunate that CP is more prevalent in people with

more limited financial resources [118,121,124,128] and endorsement of this item is unsurprising, given that 92% of the respondents reported inadequate financial resources.

6.4.3 Reliability

The ZCCS displayed adequate reliability as evidenced by the high Cronbach alpha scores at both factor and scale level; the higher the IC, the more reliable the scale [147,150,553,554]. This was unsurprising given the intermediate-strong factor loadings and high ITCs. Although four distinct factors emerged, the ZCCS displayed unidimensionality (the extent to which items are measuring the same construct). The validity of the alpha scores is dependent on the homogeneity/unidimensionality of a test, and this was substantiated through EFA [150]. The seemingly lower α scores for the family and social life domains is a testimony of the Spearman-Brown equation of prophecy which stipulates that IC is a function of test length [202-206]. The domains had the least number of items when compared to other factors. Additionally, the ZCCS also exhibited acceptable stability after four weeks given its high test-retest reliability [ICC (95% CI; 880 (0.793: 0.930)].

6.5 Study limitations

Although the study achieved the stated objectives, there were limitations. The participants were conveniently selected, and data were not screened for outliers, and this may have negatively affected the internal study validity. A lack of standardisation in data collection procedures, i.e. some questionnaires were interviewer-administered, and others were self-completed, could have affected outcomes. We did, however, try to minimise the effects of mode of outcome-administration through the research team training exercises. Additionally, all negatively worded items did not perform well during structural validation, this can imply that participants did not really understand the items, or this exposes the weakness of using bivariate correlations in item reduction [192-195].

6.6 Conclusion and recommendations

A robust Beta version of the ZCCS was produced based on structural validity which was reliable, and which resulted in the identification of four credible factors. These were physical & economic burden, concerns for the child, family relations and community participation (See Appendix 11.22 Page 336 30). Further, the preliminary analysis also supports the reliability of the ZCCS. However, before the final definitive version could be produced, there was a need for further refinement of the psychometric properties of the Beta version using Rasch modelling, evaluation of other psychometric properties such as responsiveness and construct validity and establishment of cut-off points using latent profiling analysis.

7 Chapter 7: Validation of the MSPSS structural validity and reliability evaluation

7.1 Introduction

As it was necessary to have a validated instrument with which to test the divergent validity of the ZCCS, a social support (SS) scale, the Multidimensional Perceived Social Support Scale (MSPSS) was identified, translated, adapted, and validated within the Zimbabwean context. Further, it was especially important to translate the MSPSS given the importance of the buffering effect of SS on caregivers' mental health, and we did not come across a valid SS outcome suitable for use in the Zimbabwean context.

7.2 Importance of social support

Social support (SS) is an essential buffer to stressful life events [3,54,209,512-521]. An adequate amount of SS improves mental health by mitigating the effects of negative psychosocial outcomes such as depression, anxiety, low self-efficacy, stress and loneliness or social isolation [131,512-518,555]. Further, SS is a multidimensional, latent variable that depends upon an individual's politico-social environment, socialization process and personal values/ethos amongst other factors [512,513,519-521,556,557]. The conceptualization and perception of SS are both complex and diverse, as testified by a plethora of conceptual frameworks and definitions which have been postulated to describe this subjective and yet important phenomenon [3,131,132,521]. Social support can be defined as the amount of assistance one gets through interactions with other people [131,132]. The support can be either emotional (e.g. empathy), tangible (e.g. practical help) or informational (e.g. advice) [131-134,558]. A systematic review (SR) of the psychometric properties of the cross-cultural MSPSS translations and adaptations was undertaken and published. A synthesis of the SR findings is presented in this chapter, and the detailed methodology and results are included in Appendix 11.13 (Page 311).

7.3 A systematic review of translated versions of the MSPSS[2]

The full systematic review of the translated versions of the MSPSS has been published, and a summary outline of the findings are presented here. The full paper is included in Appendix 23.

7.3.1 Introduction and rationale

Various outcome measures have been developed to measure SS [513,520]. Created initially to measure SS in American adolescents, the MSPSS has evolved as one of the most extensively utilized SS outcome measure [54,513,516,517,520,521,557]. The MSPSS contains 12 items that measure the perceived adequacy of the available amount of SS [15,559,560] (See Appendix 11.14 – Page 326). It measures the amount of SS an individual receives from three sources, i.e. friends, family and significant others/special persons. The amount of SS is rated on a seven-point Likert scale, with responses ranging from very strongly disagree (=1) to very strongly agree (=7). The cumulative/total scores range from 12 to 84. As no item response theory calibration has been applied to the tool, the scores are interpreted as the higher the score, the more significant the amount of available SS [559]. The original version of the MSPSS yielded a three-factor structure, high internal consistency (α=0.88), stability (yielded α=0.85 after three months from initial administration) and moderate construct validity as the SS scores were negatively correlated to anxiety (r=-0.18; p<0.01) and depression scores (r=-24; p<0.01) [560].

7.3.2 Summary of the MSPSS systematic review

Social support (SS) has been identified as an essential buffer to stressful life events. Consequently, there has been a surge in the evaluation of SS as a well-being indicator. The Multidimensional Perceived Social Support Scale (MSPSS) has evolved as one of the most extensively translated and validated social support outcome measures. Due to linguistic and cultural differences, there is a need to test the psychometrics of the adapted versions. However, there is a paucity of systematic evidence of the psychometrics of adapted and translated versions of the MSPSS across settings. To understand the psychometric properties of the MSPSS for non-English speaking populations by conducting a systematic review of studies that examine the psychometric properties of non-English versions of the MSPSS (The full review was published and is included in Appendix 11.15).

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[2] This chapter has been published: Dambi J, Corten L, Chiwaridzo M, Jack H, Mlambo T, Jelsma J (2018): A systematic review of the psychometric properties of the cross-cultural translations and adaptations of the Multidimensional Perceived Social Support Scale (MSPSS). Health and Quality of Life Outcomes 16:80 https://doi.org/10.1186/s12955-018-0912-0 (See Appendix 11.15)

We searched Africa-Wide Information, CINAHL, Medline and PsycINFO for articles published in English on the translation and/or validation of the MSPSS. Methodological quality and quality of psychometric properties of the retrieved translations were assessed using the COSMIN checklist and a validated quality assessment criterion, respectively. The two assessments were combined to produce the best level of evidence per language/translation.

Seventy articles evaluating the MSPSS in 22 languages were retrieved, and 22 translated versions of the MSPSS were identified. The psychometric properties which were most often reported included internal consistency, test-retest reliability, structural validity and construct validity. Many of the tools did not follow a rigorous translation process, and there was sparse evidence for structural validity. The advent of evidence-based practice (EBP) and increased usage of PROMs require quality translations to ensure reliable and valid outcome measures. The retrieved MSPSS translations, therefore, need to be utilized with caution. It was also essential to assess other psychometric properties such as responsiveness, measurement error and establishment of cut-off values to increase the clinical utility and psychometric robustness of the translated versions of the MSPSS. We, therefore, recommend the development of a standardized protocol for the translation and adaptation of the MSPSS. Future translation studies should utilize the backwards-forward translation method with particular emphasis on the use of multiple translators, reconciliation of translations, a panel of expert assessment and both EFA and CFA should be performed for factorial analysis. Most translations [16/22] were not rigorously translated (only solitary backwards-forward translations were performed, reconciliation was poorly described, or were not pretested). There was weak evidence for structural validity, as confirmatory factor analysis was performed in only nine studies. Internal consistency was reported in all studies. Most attained a Cronbach's alpha of at least 0.70 against a backdrop of fair methodological quality. There was poor evidence for construct validity.

The conclusion was that there was limited evidence supporting the psychometric robustness of the translated versions of the MSPSS and, given the variability, the individual psychometrics of a translation must be considered prior to use. Responsiveness, measurement error and cut-off values should also be assessed to increase the clinical utility and psychometric robustness of the translated versions of the MSPSS. The information gathered from the systematic review was used to guide the translation and validation of the MSPSS-Shona version.

7.4 Summary of the translation process

The full report of the translation process is included in Appendix 13, and the process is summarised in this section. The full report is not included in the main body of the text as the focus of the thesis was

on the development of the ZCCS and the MSPSS was the instrument used to allow for greater understanding of the properties of the ZCCS.

7.4.1 Introduction

The developers of the MSPSS did not lay down the guidelines for the translation of their tool, thus the choice of the ISPOR guidelines. The ISPOR guidelines were deemed appropriate as they were built upon the review and consultation of 12 other standardized translation and adaptation guidelines [453]. We utilized a multiple-stage design adapted from the guidelines by the ISPOR- Translation and Cultural Adaptation Group guidelines [466]. (See Figure 7-1)

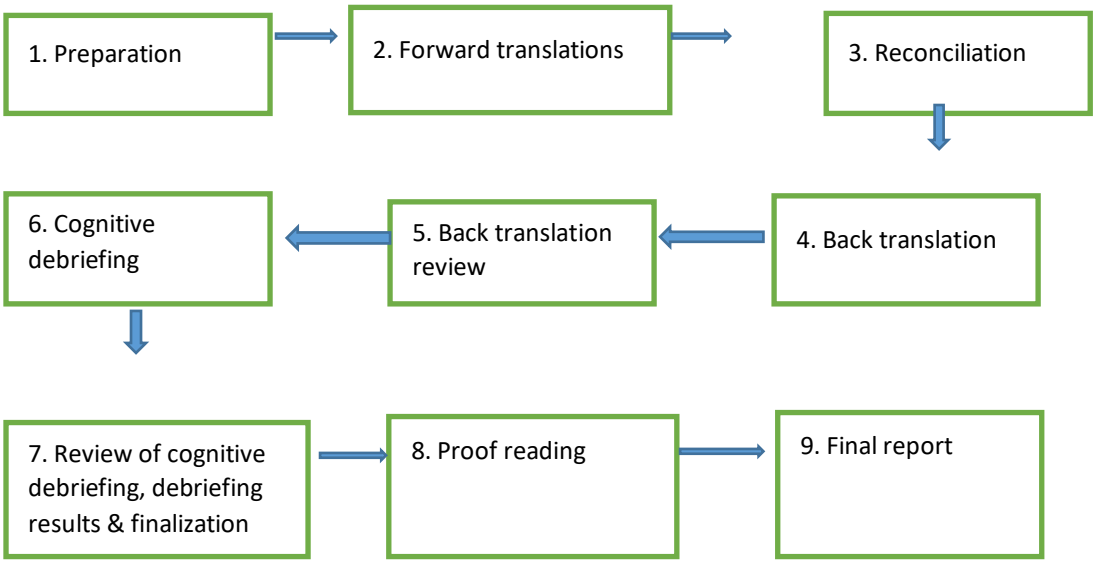


Figure 7-1: Translation process of the MSPSS

The steps that were followed are described below.

- **Preparation**

The developer granted us permission to adapt and translate the MSPSS. The developer was also invited to be involved in the translation process if clarification of any conceptual ambiguities was needed. Lastly, we recruited personnel who aided in the translation process.

- **Forward translation**

Two translators blindly translated the MSPSS into Shona. The emphasis was on attaining a colloquial translation. The translators were a Linguistics professor (T1) and a senior Linguistics lecturer (T2) from the African Languages Research Institute and University of Zimbabwe Linguistics Department respectively. T1 was not familiar with the concepts enshrined within the MSPSS and had no prior experience in translating PROMs; this was essential to get a literal translation. T2 had prior experience

in translating PROMs; this was important to ensure the attainment of a more conceptually equivalent translation. Both translators are prolific English and Shona speakers and worked independently.

- **Reconciliation**

An independent translator (T3) who was not involved in the forward translation reconciled the two forward translations into one version. T3 is a Senior Shona lecturer in the Department of Linguistics at Harare Polytechnic College. After that, there was a discussion between the two forward translators (T1 & T2), the PI, reconciliation translator (T3), and co-investigator to harmonise the reconciled Shona version.

- **Back translation**

The Shona version was then translated back to English by two independent translators (T4 & T5) who are fluent Shona and English speakers. The emphasis of the translation was on literal and conceptual meaning. The translators were senior lecturers in the Departments of English and Linguistics at the University of Zimbabwe, and both were not involved in the forward translation process. Only translator T4 had prior experience and expertise in PRO translations.

- **Back translation review**

The backward translation was compared against the original MSPSS *to ensure the conceptual equivalence of the translation* [466]*;* the PI did this with the help of the MSPSS developer.

- **Cognitive debriefing**

The translated tool was then administered to a group of caregivers of children with CP, n=16. Initially, the Shona questionnaire was administered after which the caregivers were requested to complete the English version. Respondents also evaluated the clarity of instructions, cultural relevancy, and the layout of the questionnaire.

- **Review of cognitive debriefing and finalisation**

This was done by the principal researcher with assistance from the co-investigators. This step was essential in ensuring cultural relevance by finding items which could have required modification or rewording.

- **Proofreading**

This was done to check for any typographical errors which could have been missed during the translation process. This was done by an independent linguist and a physiotherapist experienced in translation of outcome measurements who were not involved in the initial translation process to eliminate possibilities of any bias.

- **Final report**

This step involved a write up of the methodology utilized and critical decisions/amendments made during the translation process.

A robust translation process was followed, and we believe this yielded an accurate and conceptual equivalent MSPSS-Shona version. Response options were reduced from seven to five, we made use of visual prompts to simplify scoring instructions, and respondents confirmed the cultural appropriateness of the MSPSS-Shona version and were satisfied with the clarity of items and scoring instructions (See Appendix 11.13 for the detailed adaptations and amendments). However, it was important to further evaluate other psychometric properties such as structural and known group-validity and reliability by administering the survey to a large group of caregivers. More importantly, it was also essential to test the structural validity of the MSPSS-Shona version using item response theory techniques to increase the psychometric robustness of the MSPSS-Shona version further as recommended from our prior systematic review [53].

7.5 Objectives of the structural validation phase

Having previously established the face and content validity of the MSPSS-Shona version through the rigorous process of translation and cognitive debriefing of stakeholders, it was important to establish the structural validity and reliability of the outcome. The specific objectives for this phase were:

I. To apply classical test theory techniques to evaluate the factorial validity, internal consistency and test-retest reliability of the MSPSS-Shona;
II. To perform a Rasch analysis on the MSPSS-Shona to evaluate the following parameters: item and persons location, scale dimensionality, persons and item reliability indices and item invariance.

7.6 Sampling

7.6.1 Selection criterion

We selected primary caregivers who were unpaid for the caregiving role, lived with the child [540] and could understand both written and spoken English and Shona. Caregivers were excluded if they were not fluent in Shona and had a confirmed psychiatric diagnosis according to doctors' notes.

7.6.2 Sample size calculation

There are no clear guidelines for sampling estimates for CTT [155,169,179]. However, some authors recommend an optimum ratio of 5-20 candidates per item for factor analysis [155,168,169,179,541]. We set to recruit at least 400 participants since we needed two separate data sets for both exploratory

factor analysis (EFA) and confirmatory factor analysis (CFA). The pooled data set was utilized for Rasch analysis.

7.7 Instrumentation

7.7.1 Demographic questionnaire

This elicited the following caregivers' information: age, educational level, employment status, place of residence (rural vs urban), gender and perceived SES. These personal factors were important in the evaluation of item and scale invariance during Rasch analysis.

7.7.2 MSPSS

The MSPSS has 12 items and a three-factor structured SS outcome measure. Each distinct factor, i.e. family, friend and significant other/someone special, has four unique items. The original version has robust evidence of validity and reliability and does not have designated cut-off/criterial values [559-561].

7.8 Procedure

After obtaining either written or oral consent, the research team administered the MSPSS-Shona version to participants. Questionnaires were self-administered, and the research team interviewed/administered the MSPSS-Shona to caregivers who were illiterate.

7.9 Data analysis plan(s)

7.9.1 Data analysis master plan

Firstly, data were entered into Microsoft Excel and randomly split into two data sets for factor analysis. After that, we performed EFA and CFA using SPSS (Version 23), Monte Carlo Software for Parallel Analysis (Version 3) [155,169] and Stata Software (Version 15) respectively. Lastly, we ran a Rasch analysis using the RUMM2030 software.

7.9.2 Summary of structural validity testing plan

Outlined in Table 7.1 below, Table 7.2 (Page 154) and Table 7.3 (Page 154) is a summary of the EFA, CFA and Rasch analysis plans respectively:

Table 7.1: MSPSS EFA data analysis plan

Stage	Hypothesis/notes	Evaluative criteria
Testing of assumptions	Normality – data should be normally distributed	A non-significant Shapiro Wilkson Test statistic (p>0.05)
	Adequate inter-item correlations & absence of multicollinearity	I. Adequate inter-item correlations, i.e. r≥0.3 II. Items with excessively high inter-item correlations, i.e. r≥0.9 should be removed from the analysis III. Item-total correlation; ITC≥.4
	Adequate sample variation	I. KM0≥ 0.5. [KMO values are interpreted as .5-.7- mediocre, .7-.8- good, .8-.9- great & >.9 – superb] II. Participants to item ratio of at least 10:1
	Missing data	i. Missing values should occur in a non-random manner ii. Omit missing values to prevent over-estimation
	Factorability of the data	A significant Bartlett test of Sphericity- p≤ 0.05 means that the correlation matrices were statistically different from an identity matrix, i.e. there is a correlation between variables within the factor
Factor extraction method	Various factor extraction methods can be applied, and they can yield different results	i. Try out several methods and report the parsimonious solution ii. Model selection is an iterative process and should be guided by theory
Dimensionality/factor retention	Use multiple methods for factor retention	i. Kaiser's Criterion - retain factors with eigenvalues ≥ 1 ii. Horn's Parallel method - create a set of random variables, and a factor is retained if the magnitude of the random eigenvalue is less that of Kaiser eigenvalue iii. Cartel's Scree plot - inspect for breaks in continuity on the eigenvalues vs the number of factors plot. Retain all factors just before the point of inflexion
Factor rotation	Apply oblique rotation	i. Compared to Promax, direct Oblimin rotation does not allow high correlation between factors ii. Apply both methods and select the parsimonious solution
Factor interpretation	Qualitatively analyse the conceptual meaning of items loading onto an factor	i. A meaningful factor should contain at least three (3) items ii. Factor naming is an "art"

Table 7.2: MSPSS CFA data analysis plan

Fit statistic	Index	Criterion for fit
Likelihood Ratio	Chi-squared Test ($\chi ms2$)	p> 0.05
	$\chi 2/df$	<2
Population error	Root mean squared error of approximation (RMSEA)-(90% CI)	≤ 0.05
Information criteria	Akaike's information criterion (AIC)	Accept model with the lowest AIC value
	Bayesian information criterion (BIC)	Accept model with the lowest BIC value
Baseline comparison	Comparative fit index (CFI)	CFI≥0.90
	Tucker-Lewis index (LFI)	LT1≥0.90
Size of residuals	Standardized root mean squared residual (SRMR)	≤ 0.06
	The coefficient of determination (SD)	The greater the SD, the more useful the model

Table 7.3: MSPSS Rasch analysis plan

Scale trait	Method of analysis	Hypothesis/Evaluative criteria
Partial credit (PC) vs rating scale (RS) parameters	Log-likelihood ratio	If RS parameters are satisfied (ratio test is non-significant), use RS format; if not, choose PC version
Response distribution	Frequency plot of actual responses across all items and response options	Check that each possible category of item/response is endorsed; ideal to have at least 5 cases in each
Thresholds	Graphic representation and plots of probability	Ordered progression of thresholds from less to more of the trait
Person fit	Chi-square, fit residual transformed to a standardised (Z) score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all items scored by a person
Item fit	Chi-square, fit residual transformed to a Z-score	Mean 0, SD up to 1 if data fit the Rasch model; reflects the divergence between expected and actual values looking across all persons for a given item
Item trait interaction	Chi-square probability	p>0.05 tests whether the items are working as expected across the class intervals for the trait (the hierarchical ordering of items)
Individual person fit	Chi-square analysis	Values should fall within ±2.5 if the data fit the Rasch model
PSI	Cronbach's alpha	0-1; values over 0.70 allow for group comparison, over 0.85 for individual comparisons of summed scores
Local dependency	Correlation analysis of item residuals	Look for correlations of over 0.2 to 0.3
DIF	Item characteristic curves (IC curves); ANOVA of item residuals	IC curves plotted by person characteristics (such as age, sex, place of residence, SES, education) and p>0.05 for between groups ANOVA reveal DIF
Unidimensionality	Each subscale is partitioned using principal component factor analysis and subsequently t-tested	There will be no significant difference between the two partitioned pieces of the subscale

Adapted from the following sources: [187-195,207]

7.10 Study participants demographics

Most caregivers were female (98%), resided in urban areas (87.2%), were educated (97.6%), unemployed (80.3%) and reported financial challenges (64.2%) (See Table 7.4).

Table 7.4: CCWCP personal factors data

Variable	Attribute	Frequency, n (%)
Gender	Female	452 (98)
	Male	9 (2)
Place of residence	Rural	59 (12.8)
	Urban	402 (87.2)
Education	None	11 (2.4)
	Primary	83 (18.0)
	Secondary	357 (77.4)
	Tertiary	10 (2.2)
Employment	Formally employed	25 (5.4)
	Farming	19 (4.1)
	Informally employed	47 (10.2)
	Unemployed	370 (80.3)
Financial situation	Very inadequate	177 (38.4)
	Inadequate	77 (15.8)
	Neutral	177 (38.4)
	Adequate	14 (3.0)
	Very adequate	20 (4.3)

N=461

7.11 Exploratory factor analysis of the MSPSS

7.11.1 Introduction

The Maximum Likelihood, common factor analysis method, was used. Outcomes are reported in the following sequence: item descriptives, assessment of EFA testing assumptions, factor extraction, unrotated solutions, rotated solutions and finally the interpretation of factors. Results for internal consistency (IC) and test-retest reliability are then presented.

7.11.2 Item descriptives

There was a spread in the caregivers' responses on the MSPSS. Friends were cited as the least source of SS (mean 3.1 (SD 1.1)), and few participants had missing data (Table 7.5-Page 156).

Table 7.5: CCWCP descriptive statistics for MSPSS EFA analysis

Item	Subscale	Abbreviation	Mean (SD)	Lowest-Highest	Missing responses
1. There is a special person who is around when I am in need	SO	SO_need	3.3 (SD 1.5)	1-5	-
2. There is a special person with whom I can share joys and sorrows	SO	SO_joys	3.7 (SD 1.2)	1-5	-
5. I have a special person who is a real source of comfort to me	SO	SO_comf	3.7 (SD 1.2)	1-5	-
10. There is a special person in my life who cares about my feelings	SO	SO_feel	3.7 (SD 1.3)	1-5	-
3. My family really tries to help me	FAM	FAM_help	3.1 (SD 1.4)	1-5	-
4. I get the emotional help & support I need from my family	FAM	FAM_emot	3.2 (SD 1.4)	1-5	1 (.4)
8. I can talk about my problems with my family	FAM	FAM_prob	3.5 (SD 1.3)	1-5	1 (.4)
11. My family is willing to help me make decisions	FAM	FAM_deci	3.1 (SD 1.4)	1-5	-
6. My friends really try to help me	FRE	FRE_help	2.9 (SD 1.4)	1-5	-
7. I can count on my friends when things go wrong	FRE	FRE_count	2.6 (1.3)	1-5	-
9. I have friends with whom I can share my joys and sorrows	FRE	FRE_joys	3.2 (SD 1.3)	1-5	1 (.4)
12. I can talk about my problems with my friends	FRE	FRE_prob	3.1 (SD 1.3)	1-5	-
MSPSS Significant Other subscale: Range (4-20)	SO		3.8 (SD 1.0)		
MSPSS Family subscale: Range (4-20)	FAM		3.6 (SD .9)		
MSPSS Friends subscale: Range (4-20)	FRE		3.1 (SD 1.1)		
MSPSS Total: Range (12-60)	Total		42.2 (SD 10.2)		

N=231

7.11.3 Stage 1 – testing for assumptions

As shown in Table 7.6 (Page 157), data were suitable for EFA as they were normally distributed [W=.990, df=231, p=.096], exhibited adequate sampling adequacy (Kaiser-Meyer-Oklin Measure of Sampling Adequacy=.846), and the Bartlett Test of Sphericity was statistically significant (p<0.001). In addition, items correlated reasonably well with items within the same scale, there were few correlations less than 0.3, and there was no evidence of multicollinearity. The ITC range was also acceptable, i.e. 0.575 - 0.703. (See Table 7.7)

Table 7.6: MSPSS test of EFA assumptions

Attribute	Statistic	Parameters
Normality	Shapiro- Wilkinson test	W= 0.990, df=231, p=0.096
Sampling adequacy	The Kaiser-Meyer-Oklin Measure of Sampling Adequacy [KMO]	.846
Matrix identity	Bartlett Test of Sphericity	χ^2 (df 66) = 1425.7, p<0.001

N=231

Table 7.7: MSPSS inter-item correlations

Domain	**Significant other**				**Family**				**Friends**				
Items	**Item 1**	**Item 2**	**Item 5**	**Item 10**	**Item 3**	**Item 4**	**Item 8**	**Item 11**	**Item 6**	**Item 7**	**Item 9**	**Item 12**	**ITC**
Item 1	**1.00**	**0.56**	**0.59**	**0.52**	**0.48**	**0.44**	**0.26**	**0.38**	**0.22**	**0.19**	**0.11**	**0.18**	**0.65**
Item 2	**0.56**	1.00	0.61	0.52	0.50	0.40	0.32	0.33	0.27	0.25	0.20	0.22	**0.67**
Item 5	**0.59**	0.61	1.00	0.57	0.38	0.43	0.40	0.39	0.24	0.27	0.29	0.29	**0.70**
Item 10	**0.52**	0.52	0.57	1.00	0.35	0.42	0.28	0.35	0.21	0.17	0.26	0.22	**0.63**
Item 3	**0.48**	0.50	0.38	0.35	1.00	0.69	0.46	0.56	0.28	0.18	0.15	0.14	**0.68**
Item 4	**0.44**	0.40	0.43	0.42	0.69	1.00	0.50	0.59	0.28	0.09	0.20	0.15	**0.69**
Item 8	**0.26**	0.32	0.40	0.28	0.46	0.50	1.00	0.53	0.12	0.12	0.25	0.18	**0.58**
Item 11	**0.38**	0.33	0.39	0.35	0.56	0.59	0.53	1.00	0.24	0.27	0.23	0.22	**0.67**
Item 6	**0.22**	0.27	0.24	0.21	0.28	0.28	0.12	0.24	1.00	0.75	0.65	0.66	**0.64**
Item 7	**0.19**	0.25	0.27	0.17	0.18	0.09	0.12	0.27	0.75	1.00	0.64	0.68	**0.60**
Item 9	**0.11**	0.20	0.29	0.26	0.15	0.20	0.25	0.23	0.65	0.64	1.00	0.67	**0.60**
Item 12	**0.18**	0.22	0.29	0.22	0.14	0.15	0.18	0.22	0.66	0.68	0.67	1.00	**0.60**

Listwise deletion, N=231

7.11.4 Stage 2 – factor extraction

Kaiser criterion, Monto Carlo parallel analysis and inspection of the scree plot supported the retention of three factors (See Table 7.8, Table 7.9and Figure 7-2 respectively) .

Table 7.8: MSPSS Kaiser extraction criterion method

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	4.95	41.22	41.23	4.50	37.51	37.51
2	2.32	19.33	60.54	2.02	16.84	54.36
3	1.16	9.63	70.17	.71	5.95	60.31

Table 7.9: MSPSS Monte Carlo Parallel analysis extraction method

Eigenvalue #	Kaiser criterion value	***Random eigenvalue (SD)	Decision
1	4.95	1.30	Retain
2	2.32	1.22	Retain
3	1.16	1.15	Retain

*** Values for random eigenvalues are based on the following parameters: 12 variables, 231 participants and 1000 replications. A factor is retained if the magnitude of the random eigenvalue is less that of Kaiser [155,169].

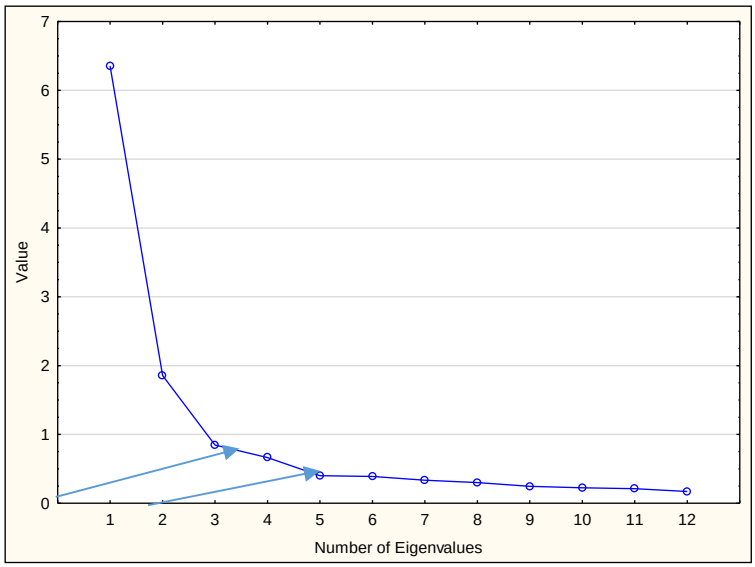


Figure 7-2: Scree plot for CWCP MPSS data

As shown in Figure 7-2 above, two inflexions at third and fifth factors support the retention of three factors.

7.11.5 Stage 3 – Factor rotation and interpretation

Three factors - friends, family and significant other - were retained upon inspection of both structure and pattern matrices (See Table 7.10 below). Factors underwent Promax (oblique) rotation method to improve interpretability. Further, item loading was high, ranging from .546 to .883. Multiple cross-loadings were especially evident for the family and significant other sub-scales.

Table 7.10: MSPSS matrices

	Pattern Matrix			Structure Matrix		
	Component			Component		
Item	**1-friends**	**2-family**	**3-significant other**	**1-friends**	**2-family**	**3-significant other**
7. FRE_count	0.88			0.86		
6. FRE_help	0.84			0.84	0.33	
12. FRE_prob	0.80			0.81		0.31
9. FRE_joys	0.77			0.77		
4. FAM_emot		0.88			0.86	0.54
3. FAM_help		0.78			0.80	0.54
11. FAM_deci		0.67			0.70	0.48
8. FAM_prob		0.55			0.60	0.43
5. SO_comf			0.83	0.33	0.50	0.82
2. SO_joys			0.72		0.51	0.75
1. SO_need			0.68		0.53	0.73
10. SO_feel			0.67		0.47	0.70

N=231

7.12 Confirmatory factor analysis

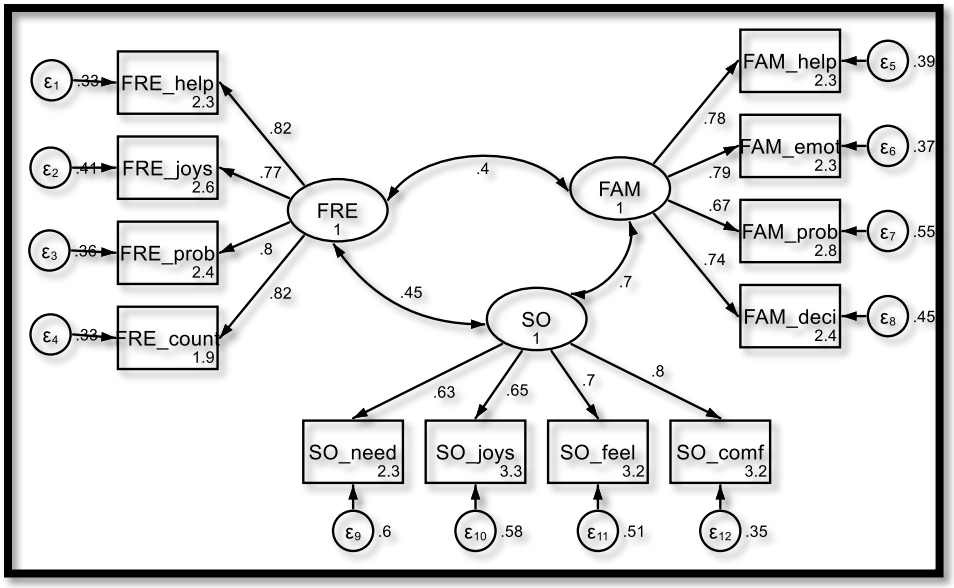
The one-factor model displayed the worst fit with the three-factor model presenting the best fit. For both the 2- and 3-factor models, there was mixed evidence for goodness-of-fit indices (gof) as the results of the chi-square test and RMSEA contradicted those of the CFI, LFI and SRMR (Table 7.11 – Page 160).

Table 7.11: MSPSS comparison of goodness of fit for the 1-, 2- and 3-factor models

Fit statistic	Index	Three-factor model	Two-factor model	One-factor model
Likelihood ratio	Chi-squared Test ($\chi ms2$)	χ2 (df 51) =147.5, p<0.001	χ2 (df 53) =233.7, p<0.001	χ2 (df 54) =564.4, p<0.001
	Standard: Interpretation	p> 0.05: poor fit	p> 0.05: poor fit	p> 0.05: poor fit
	$\chi 2/df$	2.9	4.4	10.5
	Standard: Interpretation	<2: poor fit	<2: poor fit	<2: poor fit
Population error	Root mean squared error of approximation (RMSEA)-(90% CI)	0.091 (0.074: 0.108)	0.122 (0.106: 0.138)	0.203 (0.188:0.218)
	Standard: Interpretation	≤ 0.06: poor fit	≤ 0.06: poor fit	≤ 0.06: poor fit
Information criteria	Akaike's information criterion (AIC)	8243.2: best fit	8296.1	8624.7
	Standard: Interpretation	Accept model with the lowest AIC value		
	Bayesian information criterion (BIC)	8377.5: best fit	8423.3	8748.5
	Standard: Interpretation	Accept model with the lowest BIC value		
Baseline comparison	Comparative fit index (CFI)	0.925	0.858	0.600
	Standard: Interpretation	≥0.90: good fit	≥0.90: poor fit	≥0.90: poor fit
	Tucker-Lewis index (LFI)	0.903	0.824	0.511
	Standard: Interpretation	≥0.90: good fit	≥0.90: poor fit	≥0.90: poor fit
Size of residuals	Standardized root mean squared residual (SRMR)	0.054	0.073	0.135
	Standard: Interpretation	≤ 0.06: good fit	≤ 0.06: poor fit	≤ 0.06: poor fit
	Coefficient of determination (SD)	0.993: best fit	0.981	0.876
	Standard: Interpretation	The greater the SD, the more useful the model		

N=231 Highlighted cells indicate the best fit.

As EFA supported a three-factor model, the CFA results of the model are illustrated in Figure 7-3.



FRE=Friends, FAM=Family, SO=Significant other. Abbreviations as in Table 7.5, the explanation is in the text below.

Figure 7-3: Three-factor model SEM pathway diagram

Figure 7-3 depicts the mean scores of items on the MSPSS and their correlations to one another and MSPSS subscales. The uniqueness of the specific variables is also presented. For example, for item 6 (my friends really try to help me) abbreviated FREhelp, the mean score for that item is 2.3, it loads highly/highly correlated to the friend factor (r=.82), and it uniquely contributes to 33% of the variance of the factor/domain. The higher the uniqueness, the less relevant the item is to the overall model, as uniqueness defines the amount of variance accounted/explained by that item alone [170,178].

7.13 MSPSS internal consistency

As illustrated in Table 7.12 (Page 162) and Table 7.13 (Page 162) , the MSPSS-Shona version yielded excellent IC ratings at both sub-scale and scale levels. Deletion of any of the items did not yield improvements in the scale IC thus supporting the reliability of items.

Table 7.12: MSPSS Item-total statistics, listwise deletion

Item	Scale Mean if Item Deleted	Scale Variance if Item Deleted	Corrected Item-Total Correlation	Cronbach's Alpha if Item Deleted
7. FRE_count	35.9	88.4	0.550	0.858
6. FRE_help	35.5	91.1	0.593	0.856
12. FRE_prob	35.4	89.6	0.635	0.853
9. FRE_joys	35.5	90.8	0.542	0.859
4. FAM_emot	36.1	88.0	0.595	0.855
3. FAM_help	36.0	87.8	0.601	0.855
11. FAM_deci	35.7	92.0	0.480	0.863
8. FAM_prob	36.1	89.0	0.585	0.856
5. SO_comf	36.2	89.8	0.548	0.858
2. SO_joys	36.6	91.4	0.503	0.861
1. SO_need	36.0	91.3	0.510	0.861
12. FRE_prob	36.0	91.1	0.502	0.861

N=231 *FRE=Friends, FAM=Family, SO=Significant other (Abbreviations as in Table 7.5)*

Table 7.13: MSPSS subscales IC values

Scale	SO	FRE	FAM	Scale level
α	.833	.835	.892	.868
ICC (95% CI)	.833 (.794: .865)	.892 (.868: .913)	.835 (.797: .867)	.868 (.842: .892)

N=231

7.14 Test-retest reliability of the MSPSS

The ICC (95% CI) for MSPSS scores at baseline and after four weeks was .980 (.959: 990). Data were analysed for 33 participants.

7.15 Rasch analysis for the MSPSS

7.15.1 Introduction

We utilised the Partial Credit Model (PCM), and analysis was performed using the following sequence:

- Initial model fit
- Scale targeting
- Item fit
- Threshold ordering
- Local dependency
- Unidimensionality
- DIF evaluation
- Threshold rescoring
- First scale repair analysis
- Stepwise deletion of misfitting items
- Second scale repair analysis

7.15.2 Model selection

A Likelihood Ratio Test (LRT) was performed to determine the appropriate model of analysis, i.e. PCM (partial credit model) vs RSM (rating scale model); the PCM model is the default in RUMM2030 software [188]. The LTR was statistically significant; X^2 (df 32) = 76.1, p<.001 suggesting the appropriateness of the PCM for analysis.

7.15.3 Scale targeting

Records for 461 participants were entered; of these, three (3) had missing information, and twelve (12) participants had extreme scores thus giving a final sample size of 446 participants. Figure 7-4 (Page 164)shows that items displayed a normal distribution curve with most items located around the zero logits mark. The persons mean was 0.121 implying that respondents exhibited greater trait level (SS levels) when compared to item mean. Further, the scale was well targeted as items were of varying difficulty.

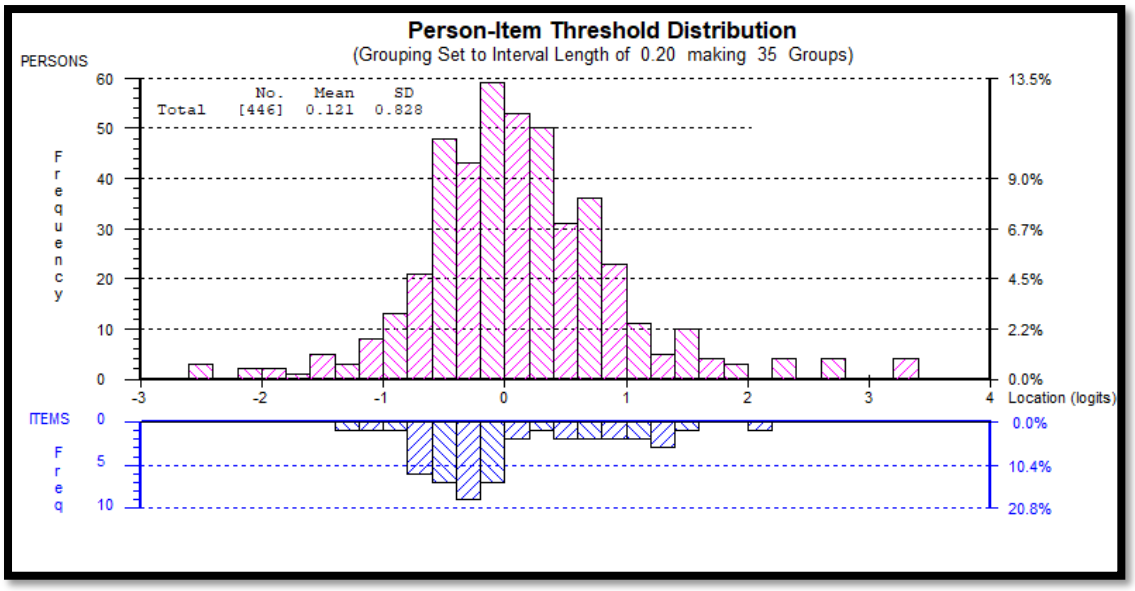


Figure 7-4: Person-item frequency distribution

7.15.4 Initial model fit

Preliminary analysis shows model misfit as the item-trait interaction was statistically significant [X^2 (df 72) = 129.1, p<.001]. However, the person separation index (PSI) and Cronbach alpha (α) were 0.873 and 0.878 respectively implying perfect targeting and good scale reliability. Further, the persons mean was higher than item mean which implies that participants exhibited greater trait level and thus understood the items (See Table 7.14 below). The RMSEA was 0.042 again demonstrating inadequate scale fit; the criterion value was RMSEA ≤ 0.003 [188].

Table 7.14: MSPSS Rasch model fit

	Items		Persons	
	Location	**Fit Residual**	**Location**	**Fit Residual**
Mean	0.0000	0.377	0.121	-0.554
SD	0.307	1.538	0.828	1.876
Skewness	0.575	-0.133	0.562	-0.670
Kurtosis	1.011	-1.497	2.387	-0.035
Correlation	[locn/std Residual]	0.513	0.035	

N=446

7.15.5 Item fit assessment

As shown in Table 7.15 below, items were of varying difficulty. Items 6 and 7 were the most difficulty with items 2 and 10 being the least difficult items. Additionally, items 5 and 8 [probabilities flagged] displayed model misfit, and their p-values were less than the Bonferroni-corrected adjusted p-value of 0.00385. None of the items displayed residuals more than the criteria value of ± 2.5.

Table 7.15: MSPSS Item fit assessment

Item	Item description	Location	SE	FitResid	DF	ChiSq	Prob	F-stat	Prob
I0001	SO_need	-0.05	0.04	1.79	404.89	9.93	0.13	1.60	0.145
I0002	SO_joys	-0.41	0.05	-0.57	404.89	6.64	0.36	1.34	0.240
I0003	FAM_help	0.11	0.04	-0.53	404.89	7.89	0.25	1.58	0.151
I0004	FAM_emot	0.07	0.04	-0.97	403.98	11.07	0.09	2.46	0.024
I0005	SO_comf	-0.32	0.05	-2.36	403.98	22.03	0.00	5.20	0.000
I0006	FRE_help	0.21	0.05	0.40	404.89	6.71	0.35	1.16	0.324
I0007	FRE_count	0.68	0.05	1.87	404.89	6.57	0.36	1.11	0.354
I0008	FAM_prob	-0.20	0.05	1.74	403.98	19.56	0.00	3.37	0.003
I0009	FRE_joys	0.10	0.05	1.92	403.98	9.94	0.13	1.67	0.128
I0010	SO_feel	-0.40	0.05	-0.96	404.89	6.94	0.33	1.54	0.164
I0011	FAM_deci	0.10	0.05	-0.27	404.89	14.04	0.03	2.73	0.013
I0012	FRE_prob	0.14	0.05	2.46	404.89	7.81	0.25	1.32	0.248

***N=231**: F statistic: Degrees of freedom 1 =6, Degrees of freedom 2 =439. FRE=Friends, FAM=Family, SO=Significant other. Abbreviations as in Table 7.5. Explanation in the text below*

7.15.6 Items threshold ordering

As shown in Table 7.16 (Page 166), only three (3) items exhibited ordered thresholds. This implies that participants could not consistently endorse or smoothly transition between the five (5) categories on most items (n=9) on the MSPSS- Shona and this is a breach of the Rasch model. The threshold map is graphically illustrated in Figure 7-5 (Page 166).

Table 7.16 : MSPSS initial threshold parameters

Item Code	Location	Mean	UnCThr 1	ThrSE 1	UnCThr 2	ThrSE 2		UnCThr 3	ThrSE 3	UnCThr 4	ThrSE 4
I0001	-0.05	-0.05	0.09	0.15	-0.47	0.13		-0.42	0.11	0.60	**0.13**
I0002	**-0.41**	-0.41	-1.02	0.16	-0.75	0.13		-0.74	0.10	0.87	0.12
I0003	**0.11**	0.11	-0.27	0.13	-0.16	0.13		-0.29	0.11	1.15	0.14
I0004	**0.07**	0.07	-0.29	0.13	-0.26	0.12		-0.02	0.11	0.84	0.14
I0005	**-0.32**	-0.32	-0.51	0.16	-0.62	0.14		-0.89	0.10	0.75	0.12
I0006	**0.21**	0.21	-0.20	0.13	-0.30	0.12		0.01	0.11	1.31	0.16
I0007	**0.68**	0.68	-0.01	0.11	0.23	0.12		0.46	0.13	2.04	0.22
I0008	**-0.20**	-0.20	-0.65	0.15	-0.60	0.12		-0.58	0.10	1.03	0.13
I0009	**0.10**	0.10	-0.35	0.14	-0.54	0.11		-0.06	0.11	1.33	0.15
I0010	**-0.40**	-0.40	-0.70	0.16	-0.21	0.16		-1.21	0.10	0.50	0.11
I0011	**0.09**	0.09	-0.59	0.13	-0.13	0.12		-0.35	0.11	1.45	0.15
I0012	**0.13**	0.13	-0.17	0.14	-0.52	0.12		-0.10	0.11	1.32	0.15

Key: shading implies disordered thresholds; UnCThr=uncentralized thresholds [location=mean]; ThrSE= threshold standard error

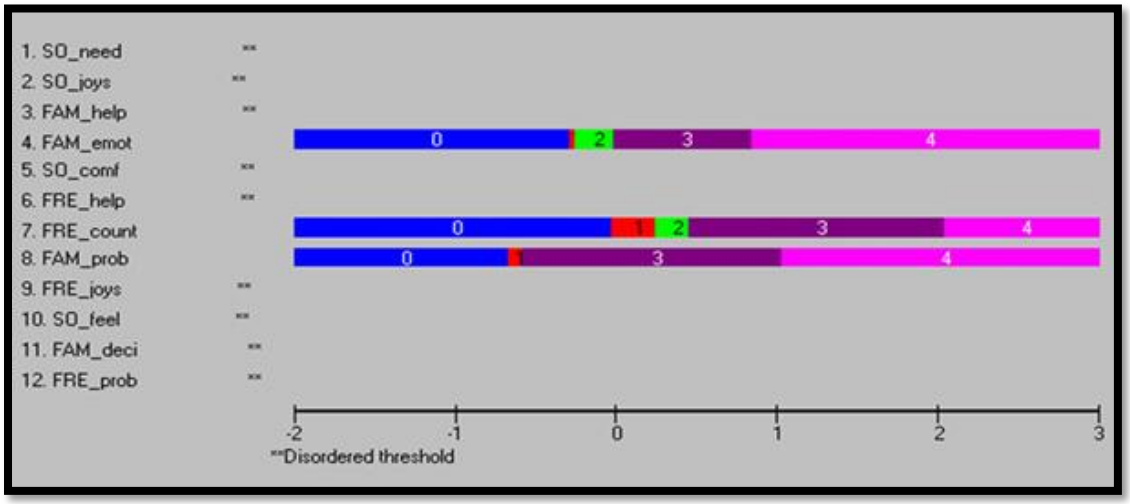


Figure 7-5: Initial threshold map

Figure 7-6 (Page 167) shows the item characteristic curve (ICC) for item 1 which had disordered thresholds. Respondents had challenges in discriminating categories: “disagree” to “neutral”. Furthermore, the threshold map further illustrates the justification of the PCM as category distances were uneven.

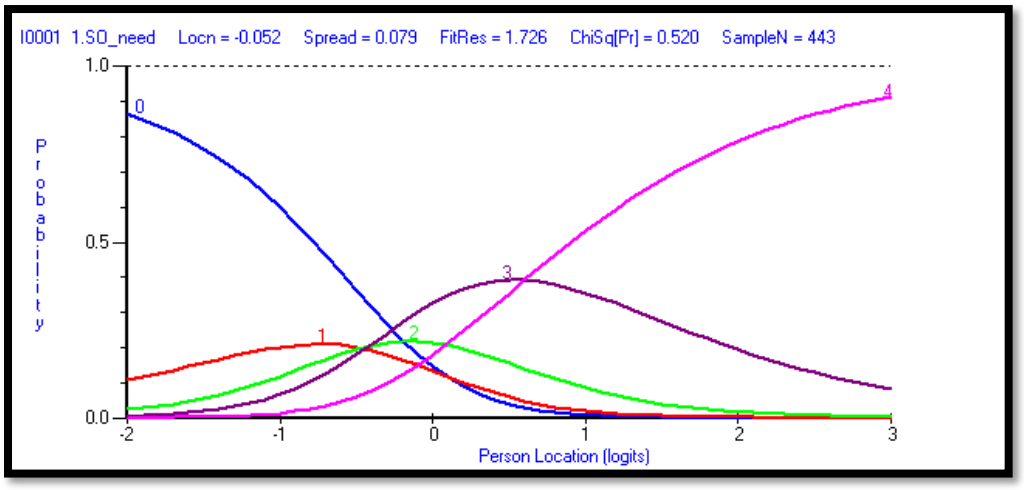


Figure 7-6: ICC for item 1

The ICC of item 4 illustrated in Figure 7-7 below illustrates an example of an ordered threshold whereby every category had the highest probability of selection/endorsement.

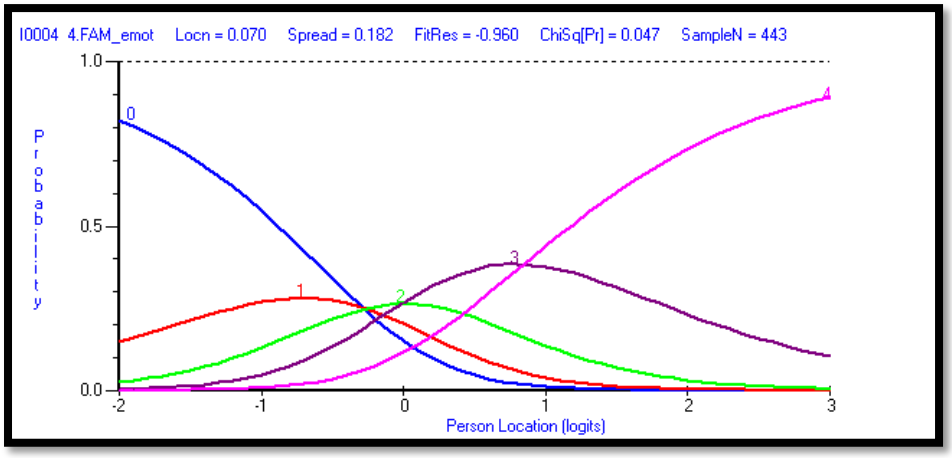


Figure 7-7: ICC for item 4

7.15.7 Local dependency

Illustrated in Table 7.17 below are the residual inter-item correlations and correlations greater than 0.1 are flagged. All items displayed local dependency which is also a breach of Rasch modelling.

Table 7.17 :MSPSS items residual correlations

Item description	Item	I0001	I0002	I0003	I0004	I0005	I0006	I0007	I0008	I0009	I0010	I0011
SO_need	I0001	1										
SO_joys	I0002	0.21	1									
FAM_help	I0003	0.08	0.13	1								
FAM_emot	I0004	-0.01	-0.08	0.45	1							
SO_comf	I0005	0.15	0.27	-0.11	-0.04	1						
FRE_help	I0006	-0.32	-0.28	-0.26	-0.26	-0.34	1					
FRE_count	I0007	-0.26	-0.26	-0.39	-0.46	-0.25	0.43	1				
FAM_prob	I0008	-0.09	-0.07	0.10	0.11	0.02	-0.35	-0.32	1			
FRE_joys	I0009	-0.40	-0.34	-0.43	-0.29	-0.28	0.40	0.30	-0.17	1		
SO_feel	I0010	0.11	0.13	-0.10	-0.05	0.25	-0.26	-0.25	-0.15	-0.15	1	
FAM_deci	I0011	-0.08	-0.13	0.15	0.21	-0.09	-0.29	-0.27	0.27	-0.23	-0.12	1
FRE_prob	I0012	-0.36	-0.30	-0.43	-0.41	-0.26	0.37	0.48	-0.30	0.42	-0.20	-0.27

Abbreviations as in Table 7.5

Further analysis revealed that there was a local dependency between items in the same domain/factor as illustrated in Table 7.18 below.

Table 7.18: MSPSS local dependency analysis

item	Domain	Local dependency	Domain-item	Domain-item	Domain-item
1. SO_need	SO	2	SO-2		
2. SO_joys	SO	1,5	SO-1	SO-5	
3. FAM_help	FAM	4	FAM-4		
4. FAM_emot	FAM	3,11	SO-3	SO-11	
5. SO_comf	SO	2,10	SO-2	SO-10	
6. FRE_help	FRE	7,9,12	FRE-7	FRE-9	FRE-12
7. FRE_count	FRE	6,9,12	FRE-6	FRE-9	FRE-12
8. FAM_prob	FAM	11	FAM-11		
9. FRE_joys	FRE	6,7,12	FRE-6	FRE-7	FRE-12
10. SO_feel	SO	5	SO-5		
11. FAM_deci	FAM	4,8	FAM-4	FAM-8	
12. FRE_prob	FRE	6,7,9	FRE-6	FRE-7	FRE-9

7.15.8 Initial unidimensionality evaluation

As shown in Table 7.19 below, principal component analysis of residuals was performed by comparing loadings on the first principal component; items were dichotomised into either positive or negative loadings based on a threshold correlation of 0.1. After extracting the Rasch factor, we contrasted high positive and high negative loadings on the first principal component of the residuals through a paired t-test. 20.9% [93 out of 446] of the estimates were significantly different, and the lower bound of the binomial confidence interval was 18.8% which is above the criteria value of 5% [188]. This suggested that the MSPSS-Shona is multidimensional, another breach of Rasch modelling.

Table 7.19: MSPSS principal component loadings

Item	PC1	PC2	PC3	PC4	PC5	PC6	PC7	PC8	PC9	PC10	PC11	PC12
I0012	0.78	0.03	0.06	-0.02	-0.01	0.14	0.07	-0.22	0.46	-0.31	-0.08	0.05
I0007	0.75	0.07	-0.09	-0.27	0.10	0.16	0.17	-0.29	-0.18	0.41	-0.03	0.05
I0006	0.72	-0.11	-0.32	0.06	-0.08	-0.05	0.05	0.24	-0.45	-0.30	-0.11	0.05
I0009	0.71	-0.11	0.19	0.25	-0.11	-0.25	-0.23	0.35	0.17	0.22	0.26	0.05
I0010	-0.25	0.54	0.18	0.62	0.21	0.14	-0.29	-0.22	-0.17	-0.02	-0.04	0.04
I0011	-0.36	-0.52	0.35	-0.10	0.31	0.54	0.02	0.28	-0.03	-0.02	0.05	0.04
I0005	-0.37	0.56	0.34	0.05	-0.23	-0.04	0.58	0.09	-0.05	-0.05	0.18	0.04
I0008	-0.37	-0.41	0.58	-0.28	-0.18	-0.36	-0.19	-0.22	-0.15	-0.07	-0.10	0.05
I0002	-0.41	0.50	-0.17	-0.29	-0.49	0.26	-0.29	0.19	0.08	0.08	-0.18	0.04
I0001	-0.44	0.41	-0.27	-0.34	0.57	-0.32	-0.04	0.13	0.10	-0.04	0.02	0.05
I0004	-0.53	-0.47	-0.28	0.41	-0.05	-0.14	0.26	0.06	0.16	0.17	-0.33	0.05
I0003	-0.57	-0.38	-0.51	0.02	-0.19	0.07	-0.06	-0.25	-0.02	-0.07	0.41	0.04

7.15.9 Differential item functioning (DIF)

DIF is when an item functions differently when applied/administered to the same population [187-189,192,195]. The differences can be either systematic or non-systematic, and this is referred to as uniform and non-uniform DIF respectively [187-189,192,195]. Items exhibiting DIF are flagged, and next sections are descriptions of the item invariance across age and level of education.

7.15.9.1 Age

Items 10 and 9 displayed uniform and non-uniform DIF by age respectively as shown in Table 7.20 (Page 170). Figure 7-8 (Page 170) visually illustrates how SS varies across age whereby older participants were more likely to have lower trait (SS) levels.

Table 7.20: MSPSS DIF by age

	Class Interval				Age				Class Interval by age			
Item	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**
I0001	1.63	1.56	6	0.15602	0.94	0.90	3	0.43949	1.32	1.26	18	0.20795
I0002	1.14	1.36	6	0.23137	1.71	2.03	3	0.10921	1.08	1.28	18	0.19959
I0003	1.37	1.59	6	0.14753	2.48	2.88	3	0.03592	0.85	0.99	18	0.46798
I0004	2.09	2.55	6	0.01931	0.30	0.37	3	0.77292	1.23	1.51	18	0.08386
I0005	3.63	5.26	6	0.00002	0.10	0.14	3	0.93402	0.98	1.42	18	0.11574
I0006	1.09	1.17	6	0.31997	1.51	1.63	3	0.18275	0.99	1.07	18	0.38532
I0007	1.27	1.24	6	0.28443	1.21	1.18	3	0.31783	1.15	1.12	18	0.33347
I0008	3.61	3.67	6	0.00147	0.88	0.89	3	0.44495	1.45	1.48	18	0.09428
I0009	1.77	1.80	6	0.09709	0.60	0.61	3	0.60797	2.37	2.41	18	0.00112
I0010	1.34	1.67	6	0.12616	5.45	6.80	3	0.00018	0.64	0.80	18	0.69696
I0011	2.31	2.68	6	0.01449	1.79	2.07	3	0.10334	1.06	1.23	18	0.23488
I0012	1.50	1.37	6	0.22332	0.31	0.28	3	0.83818	1.09	1.00	18	0.46336

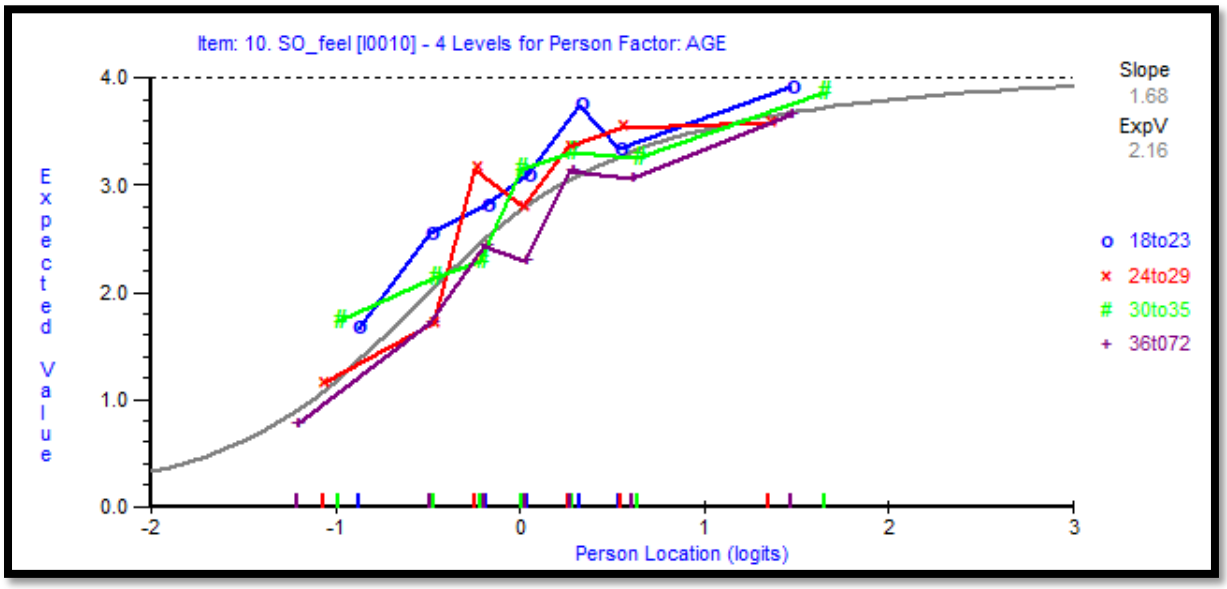


Figure 7-8: MSPSS DIF by age for item 10

7.15.9.2 Level of education

Items 12 and 9 displayed uniform and non-uniform DIF by the level of education respectively as shown in Table 7.21 below.

Table 7.21: MSPSS DIF by the level of education

	Class Interval				Education				Class Interval by education			
Item	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**
I0001	1.71	1.68	6	0.12553	3.88	3.80	3	0.01041	1.42	1.39	11	0.17442
I0002	1.08	1.29	6	0.26216	2.42	2.89	3	0.03517	1.38	1.64	11	0.08383
I0003	1.34	1.63	6	0.13672	3.80	4.64	3	0.00334	1.69	2.06	11	0.02194
I0004	1.98	2.44	6	0.02511	1.79	2.20	3	0.08784	1.25	1.54	11	0.11497
I0005	3.69	5.56	6	0.00002	1.41	2.13	3	0.09528	1.85	2.79	11	0.00159
I0006	1.12	1.23	6	0.29012	0.96	1.05	3	0.37128	1.94	2.13	11	0.01728
I0007	1.23	1.23	6	0.28975	3.26	3.26	3	0.02162	2.10	2.10	11	0.01926
I0008	3.23	3.31	6	0.00342	2.89	2.96	3	0.03211	1.71	1.75	11	0.05999
I0009	1.76	1.80	6	0.09784	2.84	2.90	3	0.03474	2.90	2.96	11	0.00085
I0010	1.30	1.59	6	0.14903	2.96	3.60	3	0.01357	0.60	0.73	11	0.71087
I0011	2.36	2.76	6	0.01215	1.40	1.64	3	0.18052	1.13	1.32	11	0.20758
I0012	1.42	1.40	6	0.21427	5.88	5.77	3	0.00072	2.27	2.23	11	0.01244

7.15.10 Scale repair

7.15.10.1 Dealing with disordered thresholds

Thresholds rescoring is an iterative process; we, therefore, evaluated various rescoring options and documented changes in the model and item fit. The rescoring structure denotes the scoring labels adopted, for example, a 00011 structure presents binary response options. Before the rescoring, the original scoring structure will be 01234 denoting the adjunct Likert scaling which ranges from strongly disagree=0 to strongly agree=4. In Rasch analysis, the first class is always ascribed a zero category, and various permutations are tried out until a parsimonious solution is reached. As illustrated in Table 7.22 (Page 172), dichotomization of responses produced the best fit, the threshold map is illustrated in Figure 7-9 (Page 172). However, the scale still displayed misfit, was slightly off-target, showed local dependency and DIF which further necessitated stepwise deletion of misfitting items to improve model fit.

Table 7.22: MSPSS threshold rescoring

Rescoring structure	Analysis Power	Overall model fit - value	Item location	Persons location	Targeting	Item misfit	Uni-dimensionality	Local dependency	DIF
00012	Good	p<.001	0.00 (SD 0.511)	-0.555 (SD 1.111)	Off target-floor effects	Yes - 4	Multidimensional 14.9% [59/404]	Yes – 7 items	Yes – age & education
00011	Reasonable	p=0.013	00 (SD .758)	0.02 (1.17)	Slightly off target	None	Unidimensional 4.86% [18/370]	Yes – 6 items	Yes – age
01123	Excellent	p<.001	.00 (SD .400)	0.07 (SD 1.0335)	Targeted - almost	Yes – 1	Multidimensional – 19.73% [88/446]	Yes -11 items	Yes – age, education

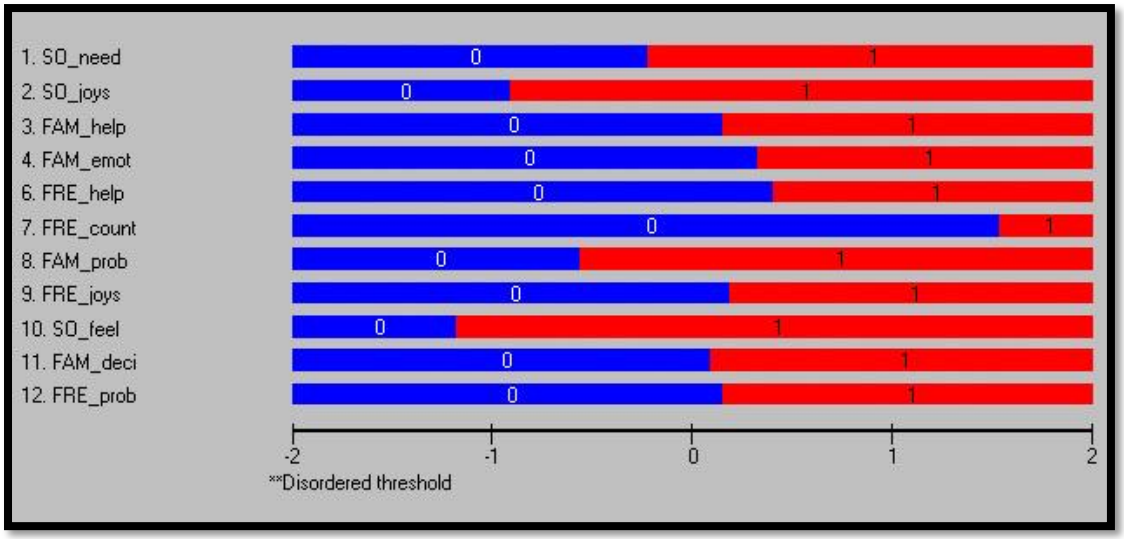


Figure 7-9: MSPSS threshold map after item 5 deletion

7.15.10.2 Item stepwise deletion

Deletion of item 5 ("I have a special person who is a real source of comfort to me.") resulted in improvement of model parameters, and the preceding sections are an outline of the scale repair analysis.

7.15.10.2.1 Scale targeting

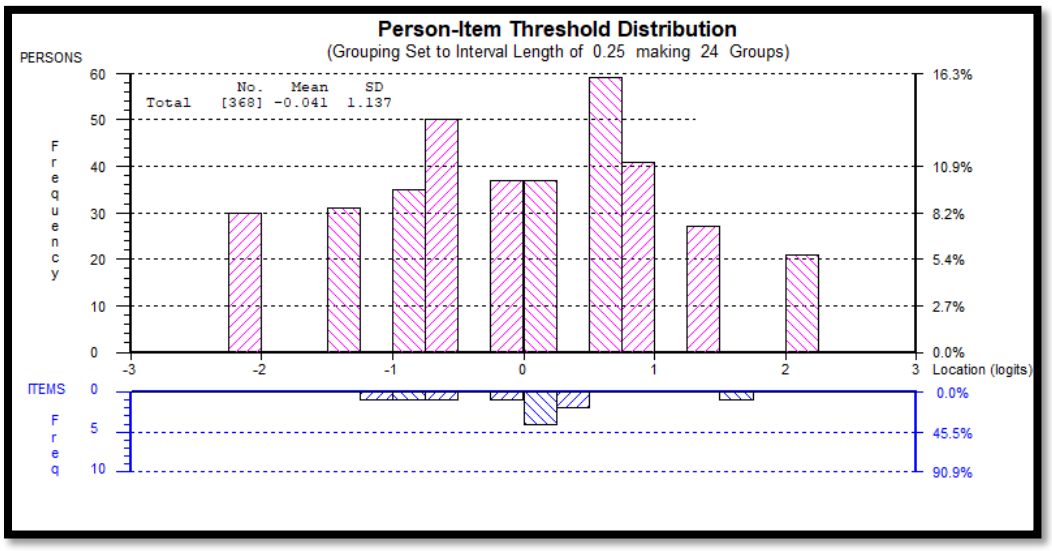


Figure 7-10: MSPSS person-item frequency distribution after item 5 deletion

Figure 7-10 above shows that the scale was well targeted as there was an equivalent number of participants with either high or low trait levels. Further, item difficulty was well spread although the distribution displayed slight deviation from a normal distribution, and peaks were observed on either side of the zero-logit landmark [mean -0.04 (SD 1.1)].

7.15.11 Revised model fit

Deletion of item 5 resulted in model fit, and the item-trait interaction was statistically non-significant [X^2 (df 66) = 81.76, p=0.091. The person separation index (PSI) and Cronbach alpha (α) decreased to 0.731 and 0.722 respectively implying perfect targeting and good scale reliability. The RMSEA was 0.001 again proving adequate scale fit (the criterion value is RMSEA ≤ 0.003 [188]).

Table 7.23: MSPSS Rasch model fit after item 5 deletion

	Items		**Persons**	
	Location	**Fit Residual**	**Location**	**Fit Residual**
Mean	0.000	0.172	-0.041	-0.031
SD	0.728	1.401	1.137	0.782
Skewness	0.376	-0.386	-0.060	0.751
Kurtosis	1.126	-1.604	-0.648	0.081
Correlation	(locn/std Resid)	-0.718	0.145	

N=368. By default, Rasch modelling omits all records with missing information, and this led to a reduced sample, i.e. n=368 against the original sample size of 461 participants.

7.15.12 Item fit assessment

As shown in Table 7.24 below, items were of varying difficulty[3]. Again items 6 and 7 were the most difficulty with items 2 and 10 being the least difficult items. None of the items displayed misfit and neither was there an item with fit residuals more than the criteria value of ± 2.5. This shows that all items fit the revised model.

Table 7.24:MSPSS item fit assessment - after item 5 deletion

Item	Item description	Location	SE	FitResid	DF	ChiSq	DF2	Prob	F-stat	Prob3
I0001	1. SO_need	-0.22	0.12	1.26	333.6	3.64	6.00	0.726	0.61	0.723
I0002	2. SO_joys	-0.91	0.12	1.66	333.6	8.59	6.00	0.198	1.39	0.218
I0003	3. FAM_help	0.16	0.12	0.21	333.6	6.85	6.00	0.335	1.24	0.287
I0004	4. FAM_emot	0.33	0.12	-1.92	332.7	13.74	6.00	0.033	3.04	0.007
I0006	6. FRE_help	0.41	0.12	-1.92	333.6	12.23	6.00	0.057	2.80	0.011
I0007	7. FRE_count	1.54	0.14	-1.46	333.6	6.11	6.00	0.411	1.16	0.325
I0008	8. FAM_prob	-0.56	0.12	1.53	332.7	4.14	6.00	0.658	0.71	0.641
I0009	9. FRE_joys	0.19	0.12	0.03	332.7	4.92	6.00	0.554	0.81	0.564
I0010	10. SO_feel	-1.18	0.13	1.12	333.6	7.28	6.00	0.296	1.12	0.350
I0011	11. FAM_deci	0.09	0.12	-0.23	333.6	7.05	6.00	0.316	1.29	0.259
I0012	12. FRE_prob	0.15	0.12	1.61	333.6	7.20	6.00	0.303	0.98	0.441

F statistic degrees of freedom DF1=6; DF2=361 Shaded blocks = more "difficult" items

7.15.13 Local dependency

Illustrated in Table 7.25 (Page 175) is the residual inter-item correlations and correlations greater than 0.3 are flagged. Only item pairs 3 and 4 and 7 and 12 displayed local dependency (a breach of Rasch modelling).

[3] "The difficulty (challenge, easiness, etc.) of an item (task, prompt, etc.) is the point on the latent variable (unidimensional continuum) at which the highest (most difficulty) and lowest (easiest) categories have equal probability of being observed." https://www.winsteps.com/winman/itemdifficulty.htm Accessed 17 October 2018.

Table 7.25 : MSPSS items residual correlations after item 5 deletion

Item	I0001	I0002	I0003	I0004	I0006	I0007	I0008	I0009	I0010	I0011
I0001	**1**									
I0002	0.094	1								
I0003	0.005	-0.056	1							
I0004	-0.016	-0.075	0.3	1						
I0006	-0.202	-0.126	-0.187	-0.228	1					
I0007	-0.166	-0.155	-0.317	-0.364	0.286	1				
I0008	-0.114	-0.088	0.025	0.068	-0.227	-0.204	1			
I0009	-0.296	-0.234	-0.321	-0.265	0.155	0.194	-0.154	1		
I0010	0.094	-0.006	-0.137	-0.057	-0.261	-0.138	-0.178	-0.098	1	
I0011	-0.142	-0.154	0.131	0.133	-0.214	-0.259	0.062	-0.217	-0.092	1
I0012	-0.286	-0.225	-0.373	-0.365	0.21	0.304	-0.231	0.295	-0.138	-0.187

7.15.14 Revised scale unidimensionality evaluation

After extracting the Rasch factor, a contrast of high positive and high negative loadings on the first principal component of the residuals through a paired t-test revealed that the revised scale was unidimensional as 2.2% [8 out of 368] of the estimates were statistically significantly different. The principal component loadings are displayed in Table 7.26 below.

Table 7.26: MSPSS principal component loadings after item 5 deletion

Item	PC1	PC2	PC3	PC4	PC5	PC6	PC7	PC8	PC9	PC10	PC11
I0012	0.72	-0.12	0.16	-0.04	0.12	0.07	0.18	0.24	0.41	-0.41	0.08
I0007	0.67	0.04	-0.17	-0.02	-0.10	0.27	-0.36	0.21	0.14	0.50	0.06
I0009	0.61	-0.17	0.28	0.02	-0.14	-0.41	0.41	-0.04	-0.30	0.25	0.07
I0006	0.56	-0.14	-0.50	-0.22	0.00	0.01	-0.21	-0.45	-0.24	-0.26	0.06
I0010	-0.13	0.54	0.65	-0.20	-0.01	-0.12	-0.41	-0.05	-0.13	-0.12	0.07
I0002	-0.20	0.53	-0.39	0.36	0.54	-0.31	0.04	0.04	0.04	0.08	0.07
I0008	-0.31	-0.37	0.08	0.78	-0.31	0.07	-0.18	-0.03	-0.05	-0.12	0.07
I0001	-0.32	0.61	-0.15	-0.08	-0.38	0.44	0.38	-0.10	0.00	0.00	0.07
I0011	-0.38	-0.46	0.28	-0.11	0.54	0.45	0.11	-0.16	-0.10	0.14	0.07
I0003	-0.58	-0.28	-0.30	-0.35	-0.09	-0.10	-0.05	0.52	-0.27	-0.10	0.07
I0004	-0.59	-0.28	-0.05	-0.28	-0.18	-0.35	-0.03	-0.27	0.48	0.16	0.06

7.15.15 Differential item functioning

Only item 8 displayed non-uniform DIF by age as shown in Table 7.27 below. Older participants were more likely to have lower trait (SS) levels.

Table 7.27: MSPSSDIF by age after item 5 deletion

	Class Interval				Age				Class Interval by Age			
Item	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**	**MS**	**F**	**DF**	**Prob**
I0001	0.69	0.68	6	0.665	1.68	1.67	3	0.174	0.9	0.8	18	0.648
I0002	**1.47**	1.37	6	0.225	1.11	1.04	3	0.374	1.0	1.0	18	0.482
I0003	**1.10**	1.21	6	0.299	2.66	2.94	3	0.033	1.0	1.1	18	0.368
I0004	**2.37**	3.14	6	0.005	0.09	0.12	3	0.946	0.8	1.1	18	0.379
I0006	**2.05**	2.75	6	0.013	0.80	1.07	3	0.364	0.9	1.3	18	0.218
I0007	**0.82**	1.14	6	0.339	1.28	1.79	3	0.149	0.7	1.0	18	0.461
I0008	**0.69**	0.75	6	0.608	0.52	0.57	3	0.637	2.6	2.8	18	0.000
I0009	**0.79**	0.90	6	0.494	1.78	2.03	3	0.11	1.4	1.6	18	0.055
I0010	**1.14**	1.13	6	0.347	3.90	3.86	3	0.01	1.1	1.1	18	0.335
I0011	**1.14**	1.30	6	0.257	1.65	1.89	3	0.132	1.1	1.3	18	0.194
I0012	**1.07**	1.08	6	0.374	0.65	0.66	3	0.58	1.7	1.7	18	0.034

7.15.16 Summary of item repair

After dichotomisation of scores and deletion of item 5, the scale displayed model fit, unidimensionality, scale targeting, adequate reliability and all items showed adequate fit. However, two pairs of items still displayed local dependency and item 7 displayed non-uniform DIF by age. Taken together, the revised 11-item scale is a valid metric outcome measure.

7.16 Discussion

7.16.1 Classical testing theory

Both EFA and CFA methods replicated the original three-factor structure. This suggests that caregivers could distinguish between the three sources of SS, i.e. family, friends and significant other. The high inter-item correlations (range: 0.464-0.745) and ITC (range: 0.575 - 0.703) are further evidence that items were measuring the same construct (SS). A three-factor structure has also been replicated in Chinese caregivers of children with CP [562], and in Malawi [47,563], Uganda [3], and Nigeria [54,555,564] and these countries have a similar socio-cultural context.

Recommendations from our systematic review (SR) on the psychometric properties of translated versions of MSPSS versions [53] guided the development of a robust methodology for the translation and validation of the MSPSS-Shona version. Firstly, we utilized an adequate sample size, and this ensured maximal variation in participants' responses as shown by adequate sampling adequacy, a non-significant Bartlett Test of Sphericity and normal distribution of the data which are pre-requisites for accurate factor analysis [165,171].

Secondly, we utilized both EFA and CFA despite CFA being the preferred structural validity technique when evaluating the dimensionality of a translated tool by the COSMIN guidelines [149,150,154,455]. The use of EFA is defensible given that SS is a subject and context-specific attribute, thus the need for exploring the factorial structure [3,132,131,521]. EFA is a technique of choice when the dimensionality of a tool is unknown which was not the case with the MSPSS. However, in EFA, data are superior to the analysis model, i.e. we ran EFA for the data to give evidence of dimensionality as opposed to only testing a pre-existing structure [155-157] which is the essence of only performing CFA [168,177]. More so, we had collapsed the scoring options from seven to a five-point Likert scaling, and this might have altered the structural validity. Furthermore, the perception of SS can vary across cultures; for instance, a one-factor solution has been replicated in Asian countries such as Turkey [565-568], Thailand [569-571] and Pakistan [572-576], which are considered as "collectivistic" societies [577]. This implies that respondents could not differentiate between the support provided by family, friends and significant others as postulated by the developers which necessitated the application of both EFA and CFA in the present study.

Thirdly, the use of Kaiser criterion, inspection of the scree plot and Horn's parallel method enhanced the credibility of the factor extraction process. Although Kaiser's criterion of eigenvalues is the most popular factor extraction method [164,165], it invariably overstates the number of factors extracted. There has therefore been advocacy for usage of multiple methods for factor extraction [165,166,169]. Unlike previous studies [3,513,578], we utilized oblique rotation to improve interpretability of factors. Oblique rotation is used when factors are hypothesized to be related which is the case with the MSPSS [155,157,469].

In performing CFA, we utilized multiple goodness-of-fit indices (GOF) which is the best practice in structural equation modelling [169,177]. The evaluation of multiple models, i.e. one, two and three-factor models, further enhanced the robustness of the MSPSS-Shona structural validation [579]. As with EFA, the three-factor model yielded the best fit. However, there was mixed evidence for gof indices as the results for the chi-square test, and RMSEA were contradictory to those of the CFI, LFI and SRMR, with the former indices indicating model misfit. Given the large sample size, it was

unsurprising that the likelihood ratio (chi-square) test suggested misfit to the three-factor structure. Large sample sizes [N>200] [169,580,581] are likely to result in a significant result, and this is also similar in other studies which replicated the three-factor structure using CFA on large sample sizes [512,518,562,570,582-584]. There have been suggestions to divide the chi-value by the degrees of freedom. Using this method, a criterion of chi-square/df< 2 is deemed as an acceptable fit [580], but this again did not yield adequate fit. Nevertheless, some authors argue that a $\chi 2/df$ in the range 2-3 can be deemed as acceptable fit [178].

The lower confidence limit of the RMSEA approached the criterion value, and this suggests that the MSPSS-Shona indeed has a three-factor structure [580,581]. There is controversy in the literature regarding the optimal cut-off values for RMSEA in assessing model fit [152]. Validation of the MSPSS in Malawi [512], Arabia [582], France [518], Hausa (Nigeria) [585], Poland [583], Portugal [584] and Thailand [570] also yielded misfit of the population error index (RMSEA> 0.05). Large sample size also influences the RMSEA, and other authors have advocated that RMSEA≤ 0.10 as an acceptable fit [580,581]. Given these model deviations, it was, therefore, important to scrutinize other gof indices (which indicated model fit) in evaluating the overall model fit of the MSPSS-Shona version. Taken together, the MSPSS-Shona can be deemed to have a three-factor structure since the CFI, LFI and SRMR indices indicated model fit.

The MSPSS-Shona exhibited adequate reliability given the high Cronbach alpha scores at both factor and scale level. This was unsurprising given the strong factor loadings and high ITC. IC evaluates the homogeneity of items within a scale, i.e. it assesses if items are measuring the same construct, with the higher the IC, the more reliable the scale [147,150,553,554]. The validity of the alpha scores is dependent on the homogeneity/unidimensionality of a test, and this can be established through factor analysis [150]. Replication of the three-factor structure through EFA and CFA (the preferred unidimensionality test for translated tools) further attests to the reliability of the scale [150] [158,586]. The MSPSS-Shona also exhibited stability after two weeks given its high test-retest reliability [ICC (95% CI; 980 (.959: 990)]. However, a sub-optimal sample [N=33] is a threat to the evidence of the longitudinal validity of the MSPSS-Shona. The optimal sample size is at least 50 participants [149,455,553]. Although we replicate the original three-factor solution, our findings need to be interpreted with caution due to some methodological limitations/violations of CTT assumptions. Participants were conveniently selected, and this could have led to selection bias and as such limit the applicability of the MSPSS-Shona to other populations [168]. Additionally, data were not screened for outliers, and this might have negatively influenced the model fit estimation during CFA evaluation [169].

7.16.2 Item response theory

Preliminary analysis showed that the MSPSS-Shona did not meet the requirements of Rasch modelling. The scale exhibited multidimensionality, local dependence and disordered thresholds. However, the scale displayed perfect targeting and good scale reliability. Additionally, item 5 (being comforted by a SO) and 8 (having a family to discuss problems with) displayed model misfit. However, participants exhibited greater traits as the persons' mean was higher than that of the item mean, implying that respondents understood the concept of SS well [187,188].

On the contrary, respondents had challenges with transitioning between response options as most items [9/12] exhibited disordered thresholds. This implies that respondents could not consistently endorse response options, i.e. respondents with higher levels of SS did not consistently endorse high scoring options as expected in Rasch modelling [187,188]. The original MSPSS has a 7-point Likert response format, and during the adaptation process, we collapsed the responses to five options. We anticipated that respondents might have difficulties in discriminating the scoring options as we could not find colloquial and conceptually equivalent expressions for all seven response options in the Shona language. The Shona language has 13 dialects [587-591], and this makes harmonization of translations challenging. Disordered thresholds could also imply that the scoring instructions were not clear or the difference between "strongly disagree" and "disagree" are subtle in the Shona language. Previously studies have successfully collapsed the response options to three [582,592], four [521], five [3,47,563], and six [584] levels against the original seven-point Likert scoring system and still replicated the three-factor solution. Additionally, translations of the MSPSS in Uganda [3] and Nigeria [54,555,564] made use of visual prompts to simplify the scoring instructions. We applied the same strategy. Unfortunately, our analysis still yielded disordered thresholds. However, none of the studies evaluated the structural validity using Rasch analysis, and this makes a comparison with the present study difficult. More so, there is a debate as to the utility of a 5-point Likert scale as most participants are unlikely to select the "neutral"/ middle category and this has been speculated as a potential source of disordered thresholds [192]. On the contrary, respondents were satisfied with the clarity of scoring instructions and wording in the cognitive debriefing phase, the scale did not exhibit DIF by the level of education, and most of the study population [97.6 %] were educated. Future qualitative studies are needed to understand respondents' understanding of items on the MSPSS further.

Preliminary analysis also revealed the multidimensionality of the MSPSS-Shona, which is another breach of Rasch modelling. This can imply that the tool may be measuring SS and another construct. Item misfit, disordered thresholds and local dependence are potential sources of multidimensionality [188,192,194,197]. According to Pallant and Tennant [188], local dependency is very prevalent in

health-related PROMs. In the present study, there was local dependency between items in the same domain/factor, i.e. there was a high correlation of residuals of items within the MSPSS. The high IC, inter-item correlations as evidenced by CTT analysis may explain the local dependency of items within the same domain/factor. Initial analysis also revealed that items 10 and 9 displayed uniform and non-uniform DIF by age respectively, i.e. older participants were more likely to have lower trait (SS) levels. This is unsurprising as literature postulates that, with ageing, there is a decrease in social networks and this subsequently leads to increased loneliness and decreased SS [131,132,194,555,593,594].

As the preliminary analysis yielded model misfit, we proceeded with the scale repair. Scale repair analysis is an iterative process, and it may entail item rescoring, item deletion and removal of participants with extreme scores [187-189,191,194,207]. Dichotomization of responses produced the best fit. However, the scale still displayed misfit, was slightly off-target, exhibited local dependency and DIF which further necessitated stepwise deletion of misfitting items to improve model fit. In Rasch analysis, it is permissible for items within the same scale to have a varying number of response options. This is not so in CTT whereby items within a scale should have the same number of scoring levels [187-189,2017]. After trying several options, the dichotomisation produced a parsimonious solution. Parsimony is especially crucial if the scale is to be used for routine clinical and research use. Given that all items initially exhibited disordered thresholds, this seemed to be the best option as previous studies have elucidated that the use of multiple response options may decrease the feasibility of using a PROM [189].

Dichotomization of scoring options and the deletion of item 5 resulted in the scale attaining; model fit, unidimensionality, scale targeting (absence of floor and ceiling effects), adequate reliability and all items showed adequate fit. However, deletion of an item may compromise the content validity of a PROM [197,595]. Nevertheless, in Rasch analysis, the model is superior to the data, and the aim of the scale repair is the attainment of model fit [188,192-194]. After scale repair, two pairs of items still displayed local dependency and item 7 displayed non-uniform DIF by age. The high person and item measures reliability indices, however, offset the slight model misfit. Taken together, the revised 11-item scale is a real metric outcome measure. However, further studies are needed to assess the performance of the dichotomised 11-item scale against the original scale.

To our knowledge, this is the first attempt at evaluating the psychometrics of the MSPSS using Rasch analysis. Our analysis yielded excellent reliability indices, high power of fit due to the large sample size, and we recruited a heterogeneous sample (both urban and rural participants). However, dichotomisation of responses as a “simplistic” way of dealing with disordered thresholds might have been a potential limitation as Rasch analysis permits different response formats per item [193].

However, having a different number of response options could be potentially confusing for respondents and further presents a challenge in deriving meaningful category labels for all the items given the lack of morphological differentiation of the Shona language [193]. Secondly, given that we are still naïve in IRT, we could not perform advanced scale repair techniques such as the creation of sub-test lets/super items, anchoring of items, the omission of extreme persons this might have improved the scale performance, Rasch analysis is regarded as a "…sophisticated statistical approach…" [195]. Lastly, we could not provide a transformation table which can be used to convert scores from logits to ratio scale [193] given the breaches in Rasch modelling. There is a need for further evaluation of the psychometric properties of the MSPSS-Shona to obtain true metric scores.

7.17 Conclusion

Both psychometric analytical approaches yielded different solutions. There is a growing impetus in utilising modern psychometric evaluation methods such as Rasch analysis in increasing the psychometric robustness and clinical utility of PROMs [188,191,192,194,197,196]. The discrepancies in outcomes can be accounted for by the philosophical differences and parameter estimation of both methods [168,194]. For instance, factor analysis (CTT) yields sample-dependent and putative factor solutions [166,191,192]. It is also uncommon for PROMs developed through CTT (as with the MSPSS) to fail to meet the requirements of Rasch modelling [193,194]. Failure to meet Rasch modelling does not necessarily imply that a tool is not psychometrically robust. However, the PROM should be treated as an ordinal scale during statistical analysis, unlike the current state whereby parametric tests are applied over tools developed through CTT methodologies, which is "mathematically incorrect" [196]. Nevertheless, either solution (11- or 12-item solution) must be utilised with caution in Zimbabwean caregivers given the methodological limitations. There is, therefore, a need for further exploration of the dimensionality of the MSPSS-Shona and assessment of other psychometrics such as known-group validity, divergent validity (See Chapter Eight) and establishing cut-off points to increase both research and clinical utility.

8 Chapter 8: Further psychometric evaluation of study instruments & determination of caregiver mental health

8.1 Introduction

Although credible Beta versions of the ZCCS and MSPSS had been developed and found to have robust psychometric properties (See Chapter Six and Chapter Seven respectively), there was still a need to further test the construct validity and reliability of the study instruments. In the process of this validation, a profile emerged of the mental health of Shona-speaking caregivers in Zimbabwe. Thus, this phase aimed to evaluate the psychometric properties of the ZCCS and MSPSS further and, by so doing, to determine the well-being of caregivers of children with cerebral palsy (CCWCP).

8.2 Objectives

The specific objectives were to:

- Determine if there was a statistically significant difference between caregivers of children with CP and caregivers of children with minor health problems regarding:
    - Caregiver strain as measured by scores on the ZCCS and CSI,
    - Health-related quality of life as measured by scores on the EQ-5D,
    - Social support as measured by scores on the MSPSS,
    - The frequency of common mental disorders as measured by scores on the SSQ
- To evaluate the concurrent validity of the ZCCS by establishing if there is a correlation with CSI scores;
- To assess the predictors of caregiver burden using univariate analysis;
- To perform structural equation modelling to test the relationship between caregiver burden, social support, psychiatric morbidity, HRQoL and contextual factors.

The study was done in the rural district and provincial hospitals in Mashonaland Central and West provinces. Urban participants were recruited from Harare Central Hospital and City of Harare Polyclinics. A comparative, descriptive, analytical cross-sectional design was used.

8.3 Study population

The participants were caregivers of; children with CP, children with minor health problems, and typically developing conditions. Participants were selected based on the following criteria applied to their children:

- Children with CP:
    - Diagnosis of CP according to the patient notes

- The child had to be younger than 12 years as the dynamics of caregiving a young child with CP are different compared to caregiving a teenager/adolescent with CP (13-18 years) because of differences in demands at the various stages of development [42]
- Receiving outpatients care
- Excluded if there was another physical, neurological disability, e.g. Spina Bifida [540]

- Children with minor health problems:
  - Suffering from a minor health condition such as with colds, mild respiratory problems (coughing), mild gastrointestinal problems (e.g. stomach ache, diarrhoea), rhinitis, or minor injury (e.g. bruises, minor cuts, wounds, headaches among others according to patient notes
  - Younger than 12 years
  - Receiving outpatients care

The inclusion criterion for caregivers included:

- should be permanent residents in districts/wards under investigation
- living with the child [540]
- primary, unpaid (informal) caregiver

The exclusion criterion for caregivers was:

- being pregnant [540]
- self-report of providing care for another elderly, chronically ill or disabled relative/child [542]
- having a history of chronic illness, such as diabetes, cardiovascular disorders such as hypertension, TB, HIV/AIDS, arthritis, heart disease, asthma, low back pain [540,542] according to both self-report and doctor's notes
- having another child at home with special health care needs [540,542]
- caring for another child below the age of two years
- a confirmed psychiatric diagnosis according to doctor's notes

8.4 Sample size calculation

The sample size was calculated based on the number of items on the alpha version of the ZCCS - 33 items. Assuming a 1:10 items to participants for EFA data reduction, the minimum sample was 330 participants per group, and oversampling was done to cater for missing responses and extreme scores.

8.5 Sampling

Caregivers residing in urban areas were conveniently selected from Harare metropolitan province. Caregivers of children with CP were recruited from the Harare Central Hospital Children Rehabilitation

Unit (CRU) and at peri-urban outreach sites serviced by the CRU. Multi-stage sampling was used to recruit caregivers living in rural areas. Two provinces were randomly selected, and these were further stratified with each district representing a stratum. Districts (8) with the highest number of cases of children with CP were then purposively chosen for data collection. Caregivers were recruited over six consecutive months. (See Section 8.6 for details of the recruitment process).

8.6 Data collection tools

8.6.1 Self-designed questionnaire

This tool was designed to elicit personal factors/demographic characteristics which were likely to affect caregivers' mental health (See Appendix 11.15.1 - Page 327). Variables elicited included: gender, age, educational level, employment status, place of residence (rural vs urban) and perceived levels of income. The factors were identified from the state-of-art literature review as described in Chapter Two.

8.6.2 The EQ-5D

The EQ-5D is one of the most commonly used generic HRQoL outcome measurement in adults [596]. It measures HRQoL in five domains, i.e. mobility, self-care, usual activities, anxiety/depression and pain/discomfort. Respondents also rate their overall health on a visual analogue scale. The ordinal responses can be converted to true numeric scores based on a societal preferences-based weighting system. The Zimbabwean utility scores were used for the present study. The EQ-5D has been validated in the Zimbabwean context, has been translated into Shona and Zimbabwean utility weights are available. The Shona version of the EQ-5D has been found to be a valid and reliable tool for measuring HRQoL in the Zimbabwean adult population [597]. ((See Appendices 11.17 (Page 329 and 11.18 (Page 331) respectively)).

8.6.3 Caregiver Strain Index (CSI)

Originally developed in the USA, the CSI is a validated and reliable tool for measuring the burden of caregiving adult patients [598]. Respondents are required to respond with a Yes or a No. A 'yes' is given a score of one and a 'no' a score of zero. Summation of yes responses gives the total score that ranges from zero to twelve. A score of seven or more signifies a high level of caregiver burden [238]. The tool has been previously validated in CCWCP in the Zimbabwean context [12]. It has evidence of construct validity as scores were negatively co-related to HRQoL outcomes (Spearman's rho = –0.33, p = 0.027). The CSI also displayed stability as there was a strong, positive correlation in scores at baseline and after three months and displayed homogeneity (Cronbach's alpha 0.80) [12,28]. See

Appendices 11.19 (Page 333) and 11.20 (Page 334)) for the English and Shona version of the CSI respectively.

8.6.4 Shona symptoms questionnaire (SSQ)

The SSQ is a 14-item tool for the screening of common mental disorders [599,600]. Respondents indicate whether they have experienced any of the 14 symptoms enlisted on the tool over the past seven days. A reported symptom ('yes' response) is rated as one and 'no' symptom reported is ranked as zero. A score of eight is regarded as a case exhibiting psychiatric morbidity [599,600]. Developed in Zimbabwe, the SSQ has demonstrated psychometric properties and has been adopted and validated in other settings [601,602]. (See Appendix 11.21 - Page 335).

8.6.5 Multidimensional Scale of Perceived Social Support (MSPSS)

The MSPSS is one of the most extensively used and psychometric sound tools in measuring social support [51]. Respondents rate the amount of support they receive from significant others, family and friends [47]. The MSPSS has demonstrated trans-cultural validity and has been translated into several African native languages in Malawi [47], Uganda [3] and Nigeria [54]. The adapted 5-point Likert scale was used for the present study. It was translated into Shona using a rigour forward and backward translation process, as described in Appendix 11.13 (Page 311). When tested within the Zimbabwean context, the tool yielded excellent reliability indices across a heterogeneous sample of both urban and rural participants. Both English and Shona versions of the MSPSS were utilised, and language selection was based on participants' preferences.

8.6.6 Zimbabwean Caregivers Challenges Scale (ZCCS)

The Beta version of the ZCCS is a 33-item caregiver burden outcome measure, which had been found to have sound psychometric properties, but its construct validity had not been tested in the target population. Responses are rated on a five-point Likert scale which ranges from strongly disagree=1 to strongly agree=5. Summative scores range from 33 to 165 and scores are interpreted, the higher the score, the higher the perceived burden of care. Preliminary analysis showed that the ZCCS has a four-factor structure, has high IC (α=0.903) and has demonstrated evidence of longitudinal reliability (ICC (CI); 0.880 (0.793: 0.930) (See Appendix 11.22 Page 336).

8.7 Procedure

Ethical approval for the study was granted by the University of Cape Town Ethical Review Board (Ref: 122/2016), Harare Central Hospital Ethics Committee (Ref: HCHEC 110316/18), City of Harare Health Services, and provincial medical directors of Mashonaland West and Central provinces and the Medical Research Council of Zimbabwe (Ref: MRCZ/A/2072). Finally, written consent was sought from the caregivers before data collection.

Participants were recruited as they presented to rural district hospital departments for services. For both groups, we placed flyers four weeks before the commencement of data collection to publicise the study to prospective participants. Flyers were placed within the outpatients and rehabilitation departments, and the research assistants based in the departments were available to attend to potential participants.

Research assistants explained the rationale of the study to the prospective participants as they presented for services. Caregivers meeting the selection criterion were requested to sign consent forms for participation. After that, the research assistants distributed the EQ-5D, ZCCS, SSQ, MSPSS and CSI questionnaires. The questionnaires were administered in a quiet, private room in the respective departments. Both English and Shona questionnaires were self-administered and research assistants interviewer-administered the tools to caregivers who were illiterate.

8.8 Data management

The raw data were de-identified by assignment of participant identification numbers. Files containing the raw data were kept in a locked drawer at the University of Zimbabwe, Department of Rehabilitation. The data were entered into Microsoft Excel and stored on a password encrypted laptop and backed up on Dropbox and iCloud.

8.9 Data analysis plan

8.9.1 General statistical plan

Descriptive statistics including; proportions, mean (SD) and median [IQR] were used to describe the data. Chi-square, Fishers' exact and Mann-Whitney U tests were used to test the association between categorical variables and continuous data. Parametric tests, e.g. t-tests and Pearson correlation coefficient, were used to analyse continuous variables. As the sample sizes were large, the central limits theorem applied, and all numeric data were analysed using parametric statistics (See Table 8.1-Page 187). Given the disparities in the number of caregivers across the two groups, weighted means and proportions were used to compute statistical differences across the groups.

Table 8.1: Phase three data analysis plan

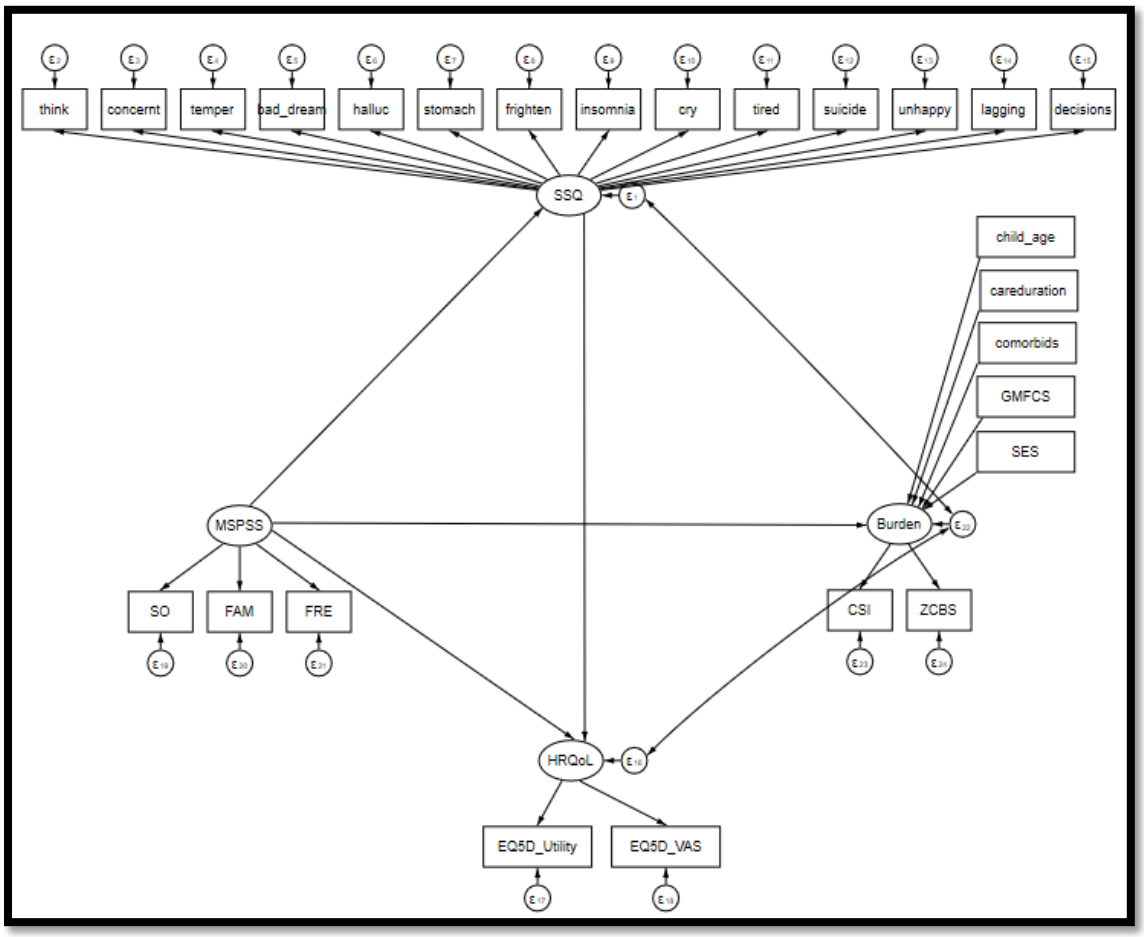
Purpose	Variable (data type)	Methods of description	Comparison statistics
Describe characteristics of children and to compare those between the TDC and children with CP	Gender (categorical)	Frequencies	Chi-square
	The severity of CP (ordinal	Frequencies	N/A
	Place of residence (nominal)	Frequencies	Chi-square
	Age (numerical)	Mean (SD)	Summary Independent samples t-tests
Describe the characteristics of caregivers and to compare differences between the two groups of carers	Place of residence(categorical) Relationship to the child (nominal) Marital status (nominal) Educational Status (ordinal) Employment status (ordinal) Perceived financial status (ordinal)	Frequencies	Chi-square
Describe the results of each outcome measure and compare between groups	EQ-5D Dimensions (ordinal) SSQ responses (nominal) CSI responses (nominal) MSPSS responses (ordinal) ZCCS responses (ordinal)	Frequencies	n/a
	Summative (numeric) indices on the EQ-5D, SSQ, CSI, MSPSS, ZCCS	Mean, SD	Summary Independent samples t-tests

8.9.2 Structural equation modelling (SEM)

Prior to applying SEM on data of caregivers of children with CP, univariate analysis was done to determine contextual factors (child- and caregiver-characteristics) which were associated with caregiver strain. Age was specifically included as the children with CP were found to be significantly older than the TD controls. Caregiver strain, SS, HRQoL and psychiatry morbidity constituted latent/exogenous factors with items on the ZCCS, CSI, MSPSS and SSQ representing observed/endogenous variables. Data were tested for normality before being entered into a specified model. The hypothesised model was based on the synthesis of the Paediatric Caregiver Model by Raina

et al. [42] and the revised Wilson and Cleary HRQoL model by Ferrans et al. [44]. As outlined in Figure 8-1 below, we tested the following hypotheses:

i. SS has buffering effects on caregiver burden and psychiatric morbidity
ii. SS optimises HRQoL
iii. Greater psychiatric morbidity is associated with higher caregiver burden
iv. Contextual factors (caregiver duration, child's age and severity of disability) influence the amount of caregiver burden



SQ=Shona Symptom Questionnaire, SO=Significant other, FAM=Family, FRE=Friends. HRQoL=Health related quality of life. CSI=Caregiver-strain Index, ZCBS=Zimbabwe Caregiver Challenge Scale, GMFCS=Gross Motor Classification System, SES=Socio-economic Status

Figure 8-1: Hypothesized caregiver mental health model

Multiple models were tested against a criterion of various gof indices, i.e. likelihood ratio: p> .05; normed chi-square; $\chi 2/df$ <2, RMSEA ≤ 0.06; CFI ≥ 0.90, LFI ≥ 0.90; SRMR ≤ 0.08 and the model with the lowest Akaike's information criterion (AIC) & Bayesian information criterion (BIC) [178-186].

8.10 Ethical considerations

- **Autonomy**

Caregivers were treated as autonomous agents and participated in the study on a voluntary basis. Caregivers either gave verbal or written consent in the presence of witnesses before participating.

- **Beneficence**

This study can potentially benefit CCWCP as study outcomes can be used to inform the development, implementation and evaluation of interventions for improving caregivers' mental health.

- **Confidentiality**

Participants were assigned a numeric code; only the researcher had access to the collected raw data which was kept in a safe locker. Electronic copies of the data were stored on a password protected laptop and backed up on Dropbox. The hard copies of the data will be shredded and burnt a year after publication of study findings.

- **Risks and benefits**

This was a relatively low-risk study; however, participants with SSQ scores ≥8 and indicated suicidal ideation were provided with counselling and support services. Where appropriate, we referred at-risk participants for further management.

8.11 Results

8.11.1 Introduction

The demographic details of the participants are presented first followed by outcomes on validation, univariate analysis and finally SEM output.

8.11.2 Socio-demographics

8.11.2.1 Socio-demographics for children

Most of the children were male (53.1%), resided in urban areas (63%), and children with CP were significantly older than TDC. The majority of TDC (73%) presented to the clinics for vaccinations. The highest frequencies in children with CP were: a diagnosis of spastic quadriplegic CP (30.2%), greater functional limitations (36.1%) and epilepsy (30.2%) (See Table 8.2- Page 190).

Table 8.2 : Children characteristics

Variable	Attribute	CP, n=461	TDC, n=771	Total, N=1 232	Statistic	p-value
Gender	Male	279 (60.5)	375 (48.6)	654 (53.1)	*X^2 (df1) = 2.444*	.118
	Female	182 (39.5)	396 (51.4)	578 (46.9		
Age in months	Median (Q_1-Q_3)	49 (26-80)	18 (10-31)	25 (14 -48)	U=72096 Z=17.477	***<.001***
Place of residence	Urban	402 (87.2)	374 (48.5)	776 (63.0)	*X^2 (df1) =31.46*	***<.001***
	Rural	59 (12.8)	397 (51.5)	456 (37.0)		
Diagnosis/service	Unspecified	164 (35.6)	Vaccination	563 (73.0)	N/A	
	Spastic quadriplegia	139 (30.2)	Weighing	112 (14.5)		
	Spastic diplegia	80 (17.4)	Flu/cold	52 (6.7)		
	Spastic hemiplegia	45 (9.8)	Diarrhoea	17 (2.2)		
	Hypotonia	11 (2.4)	Pyrexia	14 (1.8)		
	Choreo-athetosis	11 (2.4)	Gastritis	6 (.8)		
	Ataxia	11 (2.4)	Headache	4 (.5)		
			Rash	3 (.4)		
Age at diagnosis	Median (Q_1-Q_3)	4 (2-9)				
Duration of treatment	Median (Q_1-Q_3)	37 (17-67)				
Number of limbs affected	Median (Q_1-Q_3)	4 (2-4)				
GMFCS level	1	99 (21.6)				
	2	55 (12)				
	3	93 (20.3)				
	4	91 (19.9)				
	5	120 (26.)				
Co-morbid conditions	Epilepsy	139	0			
	Visual impairment	26	1			
	Speech impairment	23	0			
	Hearing impairment	21	0			
	Heart problem	10	0			
	Flu	5	27			
	Hernia	2	0			
	Gastritis	1	22			
	Other	4	5			
Number of co-morbid conditions	Range (min.-max.)	3 (0-3)	2 (0-2)			

N=1 232

8.11.2.2 Socio-demographics for caregivers

Most caregivers were female (98%), mothers to the children (95.8%), married (90.7%), educated (97.2%) and unemployed (74.7%). Compared to caregivers of TDC, caregivers of children with CP were older, had provided care for a longer duration, and reported being of a lower SES (See Table 8.3 below).

Table 8.3: Caregivers' characteristics

Variable	Attribute	CCP, n=461	CTD, n=771	Total, n=1 232	Statistic	p-value
Gender	Female	452 (98.0)	767 (99.5)	1 219 (98.9)	X^2 (df 1) = 4.388	.036
	Male	9 (2.0)	4 (.5)	13 (1.1)		
Age	Mean (SD)	34.8 (SD 10.6)	27.9 (SD 7.2)	30.5 (9.3)	t (df 1224) = 13.48	***<.001***
Relationship to child	Mother	411 (89.2)	739 (95.8)	1 150 (93.3)	X^2= 5.465 df=3	.141
	Grandmother	29 (6.3)	13 (1.7)	42 (3.4)		
	Aunt	12 (2.6)	14 (1.8)	29 (2.3)		
	Father/grandfather	9 (2.0)	2 (.3)	11 (.8)		
Marital status	Married	353 (76.6)	699 (90.7)	1 052 (85.4)	*X^2= 62.253*	***<.001***
	Divorced	51 (11.1)	40 (5.2)	91 (7.4)		
	Widower/widow	35 (7.6)	6 (.8)	41 (3.3)		
	Never lived together	11 (2.4)	18 (2.3)	29 (2.4)		
	Single	9 (2.0)	8 (1.0)	17 (1.4)		
Educational Status	Secondary	357 (77.4)	516 (66.9)	873 (70.9)	*X^2=3.264*	.353
	Primary	83 (18)	225 (29.2)	308 (25)		
	Tertiary	10 (2.2)	15 (1.9)	25 (2)		
	None	7 (1.5)	15 (1.9)	22 (1.8)		
Employment Status	Unemployed	370 (80.3)	550 (71.3)	920 (74.7)	*X^2=6.246* df=3	.100
	Informally employed	47 (10.2)	81 (10.5)	128 (10.4)		
	Formally employed	25 (5.4)	30 (3.9)	55 (4.5)		
	Farming	19 (4.1)	110 (14.2)	129 (10.5)		
Perceived financial status	Very inadequate	177 (38.4)	106 (13.7)		*X^2= 19.743* df=4	***<.001***
	Neutral	177 (38.4)	447 (58)			
	Inadequate	73 (15.8)	156 (20.2)			
	Very adequate	20 (4.3)	4 (.5)			
	Adequate	14 (3)	58 (7.5)			

N= 1 232 CCP= Caregivers of children with cerebral palsy, CTD= Caregivers of typically developing children

8.11.3 Known group validity

We assessed known group validity by comparing differences in the levels of caregiver burden, social support, HRQoL and psychiatric morbidity between the two groups of caregivers.

8.11.3.1 CSI scores

- **Frequencies of reported problems on the CSI**

Changes in personal plans, time pressure, financial strain and being overwhelmed by the caregiving role were the most reported problems (See Table 8.4 below). Further, the caregivers of TDC reported a higher frequency of 'no' responses to all item (red shading), whereas the caregivers of CP children had a more mixed pattern of responses, although most items had a majority of 'yes' responses.

Table 8.4: Frequencies of reported problems on the CSI

Item	CCP, n=461		CTD, n=771		Total, N=1 232	
	Yes (%)	No (%)	Yes (%)	No (%)	Yes (%)	No (%)
1. sleep	47.3	52.7	21.7	78.3	31.3	68.8
2. inconvenient	58.4	41.6	14.5	85.4	30.9	69.1
3. physical strain	57	43	11.2	88.8	28.3	71.7
4. confining	56.4	43.6	13.7	86.3	29.7	70.3
5. family adjustments	46.4	53.6	18.9	81.1	29.2	70.8
6. personal plans	69.6	30.4	29.1	70.9	44.2	55.8
7. time demands	84.2	15.8	23.8	76.2	46.4	53.6
8. emotional adjustments	51.3	48.7	17.7	82.3	30.2	69.8
9. child's behaviour	54.8	45.2	15	85	29.9	70.1
10. upsetting	37.4	62.6	11.8	88.2	21.4	78.6
11. work adjustments	67.2	32.8	25.6	74.4	41.2	58.8
12. financial strain	73.3	26.7	39.5	60.5	52.2	47.8
13. overwhelmed	87.8	12.2	32.2	67.8	53.1	46.9

N= 1 232 CCP= Caregivers of children with cerebral palsy, CTD= Caregivers of typically developing children. Red shading indicates highest frequency; Green shading indicated lowest frequency.

- **Caregiver Strain Index summative scores**

As shown in Table 8.5 below, caregivers of children with CP reported greater burden when compared to caregivers of TDC, and this was statistically significant, t (df 1230) =29.474, p<.001 and this is visually depicted in Figure 8-2.

Table 8.5: Comparison of CSI total scores

	CP, n=461	**TDC, n=771**	**Statistic**	**p-value**
Weighted mean (SD)	7.9 (SD 3.4)	2.7 (SD 2.6)	t (df 780.9) =28.266	p<.001
Median [Q_1-Q_3]	9 (5-11)	2 (1-4)		
Range [Min.- Max]	13 (0-13)	12 (0-12)		

CCP= Caregivers of children with cerebral palsy, CTD= Caregivers of typically developing children

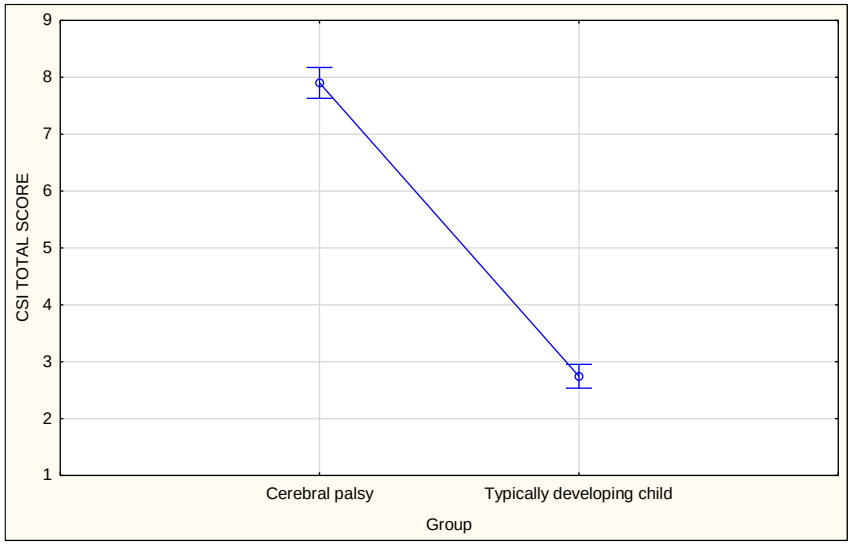


Figure 8-2: Comparison of CSI scores

8.11.3.1.1 ZCCS scores

- **Frequencies of reported problems on the Zimbabwe Caregiver Challenge Scale**

Report of bodily pain, increase in family expenses, being concerned of the child's future, poor child's functional status, and being overwhelmed by the caregiving role were the most reported problems (Table 8.6- Page 194). Visual inspection reveals that the percentages of Strongly disagree and Disagree was higher in the TDC caregivers (orange to red) and of Agree and Strongly Agree lower (green) compared to the caregivers of children with CP.

Table 8.6: Frequencies of reported levels of problem on the ZCCS

Item	Strongly disagree			Disagree			Neutral			Agree			Strongly agree		
	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %
1. time&needs	21	27	25	35	52	45	16	11	13	16	7.8	11	12	2.3	6.1
2.. future	13	20	17	33	66	54	10	4.7	6.8	24	7.7	14	20	1.9	8.9
3. tired	17	19	18	42	72	61	9.3	3	5.4	19	4.9	10	13	1.2	5.5
4. pain	13	18	16	30	68	54	7	3.8	5	31	8.2	17	19	1.9	8.1
5. phystrain	8.7	20	16	26	58	46	12	6.6	8.5	35	13	22	19	2.1	8.3
6. workadj	7.2	15	12	19	45	35	6.7	7.4	7.1	36	29	31	31	4.4	15
7. famexp	5	11	8.7	20	36	30	6.3	8.2	7.5	36	39	38	32	6.6	16
8. spefaci	5	12	9.4	16	43	33	89	10	9.7	34	27	30	37	7.4	18
9. FINstrain	4.8	12	9.4	21	48	38	10	9.4	9.7	35	27	30	29	3.5	13
10. FAM.relations	13	16	15	33	65	53	8.5	5.5	6.6	27	12	18	19	2.5	8.6
11. FAMspouse	14	18	17	32	70	56	8.5	1.8	4.3	25	7.1	14	21	2.9	9.4
12.FAMoverall	14	20	18	37	70	58	9.6	2.4	5.1	25	4.8	12	14	2.6	6.9
13. CHIbehaviour	18	17	17	41	66	57	8.7	3.8	5.6	20	11	15	12	2.2	5.8
14. CHIembrassing	13	21	18	35	63	53	5.9	3	4.1	26	11	17	20	1	8
15. SSspouse	27	17	21	39	61	53	9.2	4.2	6.1	14	12	13	12	5.5	7.9
16. PHYguilt	18	73	39	35	22	30	8.9	2.1	6.2	27	2.7	17	12	0	7.2
17. PHsleep	17	17	17	36	57	50	8	8.7	8.5	23	15	18	15	1.7	6.8
18. CHIfuture	4.3	10	8.1	6.5	42	28	4.6	6.9	6	33	33	33	51	8	24
19. CHIcry	16	20	19	34	59	50	6.3	4.4	5.1	21	14	17	22	3	10
20. CHIinsulted	11	19	16	24	57	44	5.9	4.6	5	30	18	22	30	2.3	13
21. CHIstigma	9.3	19	15	26	55	44	7	5.6	6.1	31	18	23	27	2.1	11
22. CHIcommun.	12	23	19	16	63	46	4.8	3	3.7	33	10	18	35	0.8	14
23. CHIcannot do	4.8	22	16	17	64	47	6.5	2.7	4.2	30	10	18	41	0.9	16
24. CHIsomethingb	5.2	16	12	8.9	38	27	6.3	5.1	5.5	38	36	37	42	4.8	19
25. CHIsleep	15	17	16	30	62	50	6.1	5.3	5.6	28	15	20	21	1	8.6
26. CHIill	9.5	17	14	34	65	53	9.3	4.4	6.3	25	10	16	23	2.9	10
27.CHdevelopment	8.3	20	16	22	62	47	7.4	5.7	6.3	35	10	19	28	2	12
28. PSYwitchcraft	16	28	23	25	58	46	11	2	5.5	25	10	16	22	2.1	9.6
29. PSYaccomodat	19	18	18	32	54	46	4.1	5.3	4.8	20	17	18	25	5.9	13

Item	Strongly disagree			Disagree			Neutral			Agree			Strongly agree		
	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %	CCP %	CTD %	Total %
30. SLcontact	21	27	16	35	52	55	16	11	5.3	27	7.3	15	23	1.4	9.4
31. SLhobbies	13	20	15	33	66	49	10	4.7	9	28	12	18	18	2	8.1
32. SLoverall	17	19	16	42	72	56	9.3	3	8.4	21	6.5	12	17	0.9	7
33.overwhelmed	13	18	14	30	68	49	7	3.8	11	29	8.9	17	22	3.3	10

N= 1 232 (CP, n=461 & TDC, n=771) CCP= Caregivers of children with cerebral palsy, CTD= Caregivers of typically developing children. Red shading highest frequency, Green shading lowest frequency

- **ZCCS summative scores**

Caregivers of CP reported a much higher caregiver burden when compared to caregivers of TDC. All comparisons across ZCCS subscales and ZCCS total scores were statistically significant (See Table 8.7 below). Figure 8-3 graphically further illustrates differences in ZCCS subscales scores.

Table 8.7: Comparison of ZCCS total scores

	CCP, n=461	CTD, n=771	CCP, n=461	CTD, n=771		
	Range (Q_1-Q_3)		Mean (SD)		Statistic	p-value
Physical & economic burden	4 (2.7-4)	5 (2.0-2.7)	3.3 (.9)	2.3 (.6)	t (df 707.1) = 21.2	.8
Concerns for the child	4 (2.9-4)	4 (2-2.6)	3.4 (.8)	2.3 (.6)	t (df 769.5) =25.5	***p<.001***
Family relations	4 (2.3-3.4)	4 (1.7 -2.1)	2.8 (1.1)	1.9 (.5)	t (df 575.4) =16.6	***p<.001***
Social life	4.3 (2-4)	5 (2.0-2.5)	3.0 (1.1)	2.2 (.7)	t (df 685.7) =14.0	***p<.001***
ZCCS total scores	123 (90-124)	136 (64-81)	106.1 (24.3)	72.4 (16.1)	t (df 704.2) =26.5	***p<.001***

CCP=Caregivers of children with CP, CTD = Caregivers of typically developing children

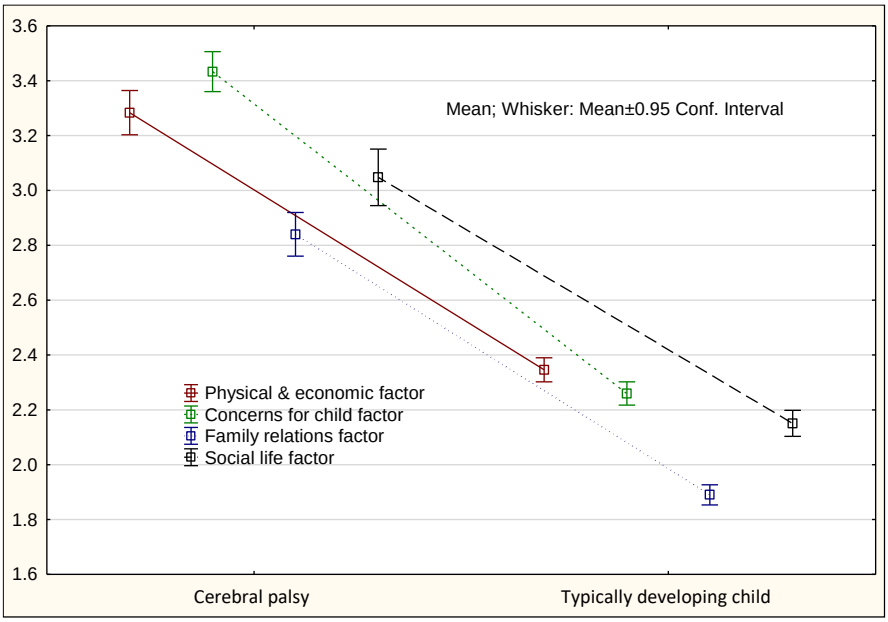


Figure 8-3: Comparison of ZCCS subscale scores

8.11.3.2 Social support

- **Frequencies of responses on the MSPSS**

Items in the significant other and friend domains were the most and least endorsed respectively (See Table 8.8 below). As can be visually seen, the caregivers of TDC reported a higher frequency of 'no' responses to all item (red shading), whereas the caregivers of CP children had a more mixed pattern of response, although most items had a majority of 'yes' responses.

Table 8.8: Frequencies of responses on the MSPSS

Item	Strongly Disagree%			Disagree %			Neutral %			Agree %			Strongly Agree%		
	CCP	CTD	Tot	CCP	CTD	Tot	CCP	CTD	Tot	CCP	CTD	Tot	CCP	TD	Tot
1. SO_need	**18**	**8.6**	12.1	**11.5**	**8.3**	9.5	**16.7**	**11**	13.2	**26.5**	**43.4**	37	**27.3**	**18**	8.6
2. SO_joys	**7.6**	6.6	7	10	6.6	7.9	17.8	9.6	12.7	36.4	48.3	43.9	28.2	7.6	6.6
3. FAM_help	**18.7**	5.3	10.3	16.3	13	14.3	17.8	16.7	17.1	26.9	43.2	37.1	20.4	18.7	5.3
4. FAM_emot	**18.3**	7.3	11.4	16.5	12.8	14.2	20	14.5	16.5	22.8	43.3	35.6	22.4	18.3	7.3
5. SO_comf	**10.7**	6.2	7.9	9.3	6.6	7.6	14.8	11	12.4	34.8	48.4	43.3	30.4	10.7	6.2
6. FRE_help	**20**	11.7	14.8	16.9	17.4	17.2	21.3	15.6	17.7	25.6	39.6	34.4	16.3	20	11.7
7. FRE_count	**29.3**	17.6	22	23.6	25.3	24.7	20	18.5	19.1	18.9	28.1	24.7	8.2	29.3	17.6
8. FAM_prob	**10.4**	5.1	7.1	12.2	9.1	10.2	18.9	13.9	15.8	34.3	49.4	43.8	24.1	10.4	5.1
9. FRE_joys	**15.9**	12.6	13.8	15	15.3	15.2	23.9	16.6	19.3	28.3	40.4	35.9	17	15.9	12.6
10. SO_feel	**10**	6.2	7.6	10.4	7.1	8.4	11.1	8.7	9.6	34.5	50	44.2	34.1	10	6.2
11. FAM_deci	**15.4**	6.9	10.1	17.8	13	14.8	18.7	16.1	17.1	30.2	45.1	39.5	18	15.4	6.9
12. FRE_prob	**17.6**	14.1	15.4	14.3	19.1	17.3	22.8	18.3	20	29.1	35.9	33.4	16.3	17.6	14.1

N=1 232 (CP, n=461 & TDC, n=771) CP=Caregivers of children with cerebral palsy, CTD=Caregivers of typically developing children, Tot=Total Red shading indicates the highest frequency; Green shading indicated the lowest frequency.

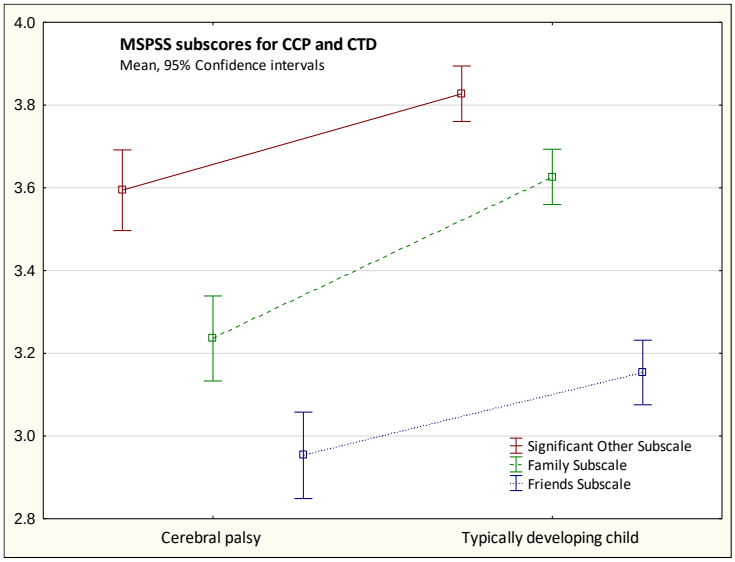
- **MSPSS summative scores**

Caregivers of TDC reported higher SS when compared to caregivers of children with CP. All comparison across MSPSS subscales and MSPSS total scores were statistically significant (Table 8.9). Figure 8-4 graphically further illustrates differences in MSPSS subscale scores.

Table 8.9: Comparison of MSPSS scores

	CCP, n=461	CTDC, n=771	CCP, n=461	CTD, n=771		
	Range [Q_1-Q_3]		Mean (SD)		Statistic	p-value
Significant other Subscale	4 (3.0-4.5)	4 (3.5-4.5)	3.6 (1.1)	3.8 (.9)	t (df 823.2) =-3.3	***p<.001***
Family Subscale	4 (2.5-4.0)	4 (3.0-4.0)	3.2 (1.1)	3.6 (.9)	t (df 823.2) =-6.60	***p<.001***
Friends Subscale	4 (2.0-4.0)	4 (2.3-4.0)	3.0 (1.1)	3.2 (1.1)	t (df 967.8) =-3.08	***p<.001***
MSPSS total score	48 (32-47)	48 (36-48)	39.1 (10.5)	42.4 (10.2)	t (df 945.7) =-5.40	***p<.001***

CCP=Caregivers of children with CP, CTD =Caregivers of Typically developing



CCP = Caregiver of children with cerebral palsy n=461, CTD= Caregivers of typical developing children n=771

Figure 8-4: Comparison of MSPSS total scores

8.11.3.3 HRQoL

- **Frequencies of reported problems on the EQ-5D**

Few caregivers had challenges in mobility, self-care and usual activities. However, caregivers of children with CP reported significantly higher levels of all dimensions, particularly of pain and anxiety/depression, when compared to caregivers of TDC (Table 8.10).

Table 8.10: Comparison of caregivers HRQoL scores

Variable	Attribute	CP, n=461	TDC, n=770	Total, n=1 231	Statistic*	p-value
Mobility	No problem (%)	73.1	92.1	85	$X^2 = 79.83$	***<.001***
	Some problem (%)	26.9	7.9	15	df=1	
Self-care	No problem (%)	93.5	97.7	96.1	$X^2 = 12.29$	***<.001***
	Some problem (%)	6.5	2.3	3.9	df=1	
Usual activities	No problem (%)	67.9	92.5	83.3	$X^2 = 126.40$	***<.001***
	Some problem (%)	27.3	5.8	13.9	df=2	
	Extreme problems (%)	4.8	1.7	2.8		
Pain/discomfort	No problem (%)	22.8	49.6	39.6	$X^2 = 137.42$	***<.001***
	Some problem (%)	49.5	43.2	45.6	df=2	
	Extreme problems (%)	27.8	7.1	14.9		
Anxiety/depression	No problem (%)	20	47.9	37.4	$X^2 = 149.51$	***<.001***
	Some problem (%)	46.6	42.5	44	df=2	
	Extreme problems (%)	33.4	9.6	18.5		

CCP = Caregiver of children with cerebral palsy, CTD= Caregivers of typical developing children

**Based on actual values, not %.*

- **Comparison of EQ-5D utility scores**

Caregivers of children with CP reported statistically significantly lower HRQoL scores when compared to caregivers of TDC (Table 8.11). Figure 8-5 and Figure 8-6 further illustrate the differences.

Table 8.11 : Comparison of caregivers' EQ-5D summative scores

		CCP, n=461	CTD, n=770	Statistic	p-value
EQ-5D utility scores	Mean (SD)	.671 (SD.206)	.834 (SD .152)	t (df 760.2) =-14.756	p<.001
	Median [Q_1-Q_3]	.727 [.525-.798]	.833 [.787-1.0]		
	Range [Min.- Max]	.997[.003-1]	.766 [.23-1]		
EQ-5D VAS scores	Mean (SD)	60.9 (SD18.9)	68.7 (SD 20.2)	t (df 1019.0) =-6.83	p<.001
	Median [Q_1-Q_3]	55 [50-70]	70 [50-90]		
	Range [Min.- Max]	90 [10-100]	96 [4-100]		

CCP = Caregiver of children with cerebral palsy, CTD= Caregivers of typical developing children

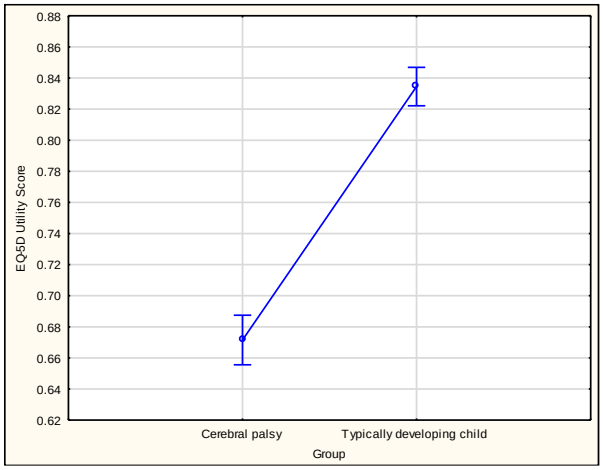


Figure 8-5: Comparison of caregivers' EQ-5D Utility scores

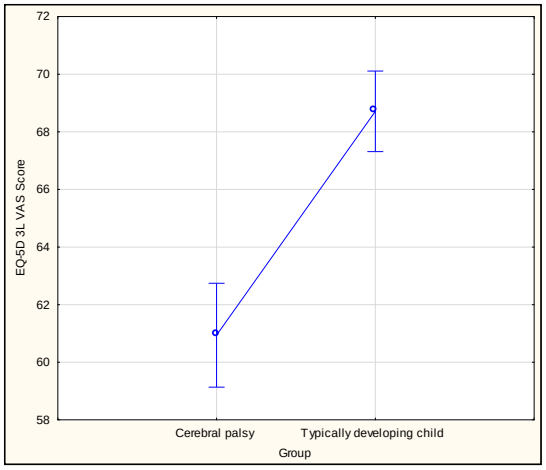


Figure 8-6: Comparison of caregivers' EQ-5D VAS scores

8.11.3.4 Psychiatric morbidity

- **Frequencies of reported problems on the SSQ**

Table 8.12: Frequencies of reported problems on the SSQ

Item	CCP, n=461		CTD, n=770		Total, N=1 232	
	Yes (%)	No (%)	Yes (%)	No (%)	Yes (%)	No (%)
1. thinking	81.1	18.9	60.2	39.8	68	32
2. concentration	75.3	24.7	54.7	45.3	62.4	37.6
3. temper	50.8	49.2	43.1	56.9	45.9	54.1
4. bad dreams	54.2	45.8	62.3	37.7	59.3	40.7
5. hallucinations	20	80	16.7	83.3	17.9	82.1
6. stomach ache	55.5	44.5	63.7	36.3	60.6	39.4
7. frightened	31.5	68.5	26.7	73.3	28.5	71.5
8. sleep	73.8	26.2	56.5	43.5	63	37
9. cry	79.8	20.2	65.4	34.6	70.8	29.2
10. tired	72.2	27.8	63.7	36.3	66.9	33.1
11. suicide	21.5	78.5	14.9	85.1	17.4	82.6
12. unhappy	52.3	47.7	35.7	64.3	41.9	58.1
13. lagging	65.1	34.9	42.5	57.5	51	49
14. decisions	71.4	28.6	56.8	43.2	62.3	37.7

CCP = Caregiver of children with cerebral palsy, CTD= Caregivers of typical developing children

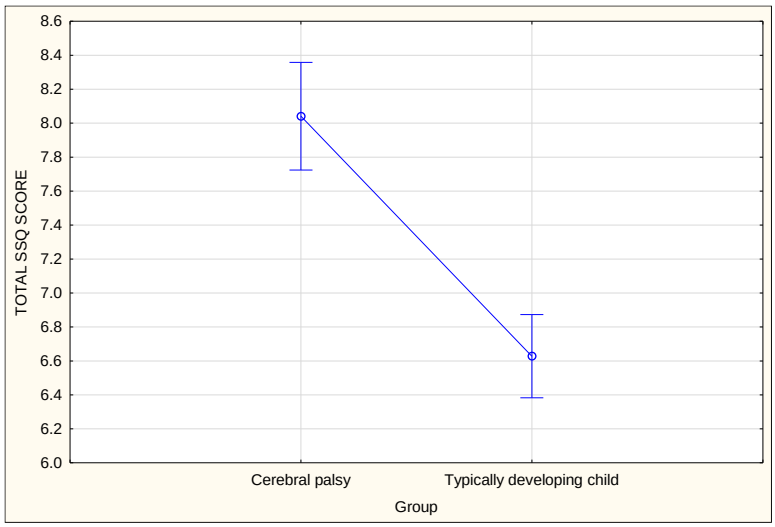
Thinking too deeply, stomach ache, feeling tired, failure to concentrate, and being overwhelmed were the most commonly reported problems (Table 8.12).

- **SSQ summative score analysis**

When compared with caregivers of CTD, caregivers of children with CP exhibited greater psychiatric morbidity (Table 8.13 below), and the differences are visually illustrated in Figure 8-7 (Page 201).

Table 8.13 : Comparison of total SSQ scores

	CP, n=461	TDC, n=770	Statistic	p-value
Mean (SD)	8.0 (SD 3.5)	6.6 (SD 3.5)	t (df 967.8) =6.794	p<.001
Median (Q_1-Q_3)	8 (5-11)	7 (4-9)		
Range (Min.- Max)	14 (0-14)	14 (0-14)		

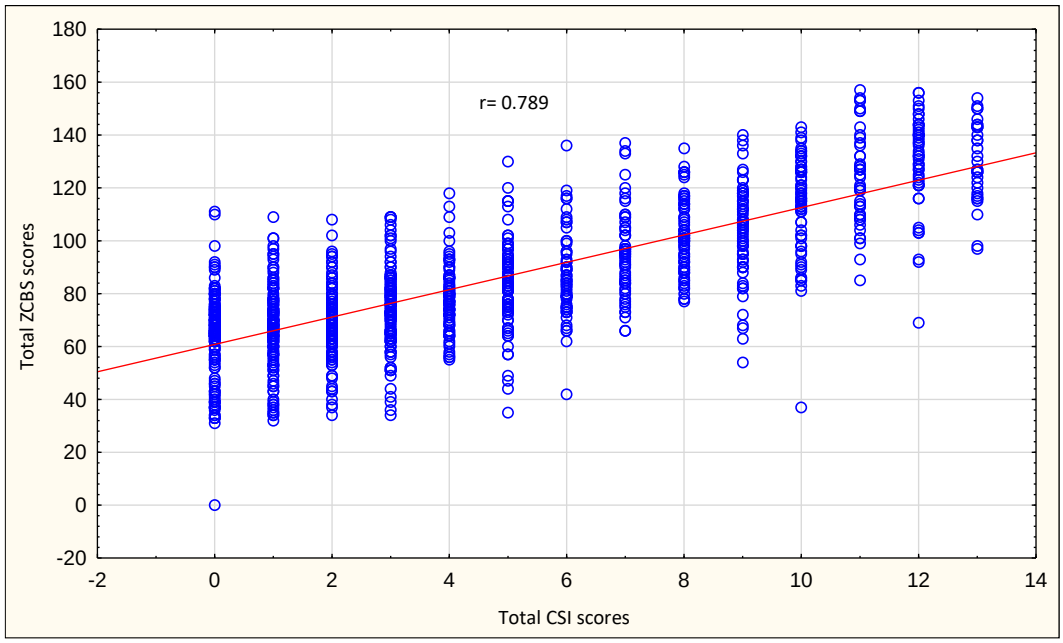


N=1,232

Figure 8-7 : Comparison of SSQ total scores

8.11.4 Concurrent validity

As shown in Figure 8-8 below, there was a strong, positive correlation between scores on the ZCCS and CSI (r=.789, p<.001), and this demonstrate the concurrent validity of the ZCCS.



N=461 Pearson's r correlation coefficient = .789, p<.001.

Figure 8-8: Relationship between ZCCS & CSI scores

8.11.5 Construct validity – hypothesis testing

As seen in Table 8.14 below, caregivers with greater caregiver burden exhibited greater psychiatric morbidity and lower HRQoL. Receiving more considerable amount of social support was associated with less caregiver burden and higher HRQoL. This is consistent with previous studies thus validating the construct (convergent) validity of the ZCCS and MSPSS. All correlations were highly significant

Table 8.14: Correlations between caregiver burden, social support, psychiatric morbidity & HRQoL

	EQ-5D Utility Score	EQ-5D 3L VAS Score	Total SSQ score	Total MSPSS score
EQ-5D Utility Score	1			
EQ-5D 3L VAS Score	.395,	1		
TOTAL SSQ SCORE	-.432,	-.400,	1	
Total MSPSS score	.339,	.283,	-.310,	
Total ZCCS score	-.527,	-.288,	.435,	-.334,

N=431: All Pearson's correlations were significant at the p<.001 level.

8.11.6 Determinants of caregiver mental health

8.11.6.1 Univariate analyses of determinants of mental health of caregivers of children with CP

Increased child age, increased caregiving duration, lower child functional capacity/more severe CP, lower education status, being unemployed, residing in urban areas and lower SES were associated with poor mental health functioning. Statistically significant outcomes are flagged, and the interpretation is provided in brackets (See Table 8.15).

Table 8.15: Determinants of the mental health of caregivers of children with CP

Variable	EQ-5D- Utility (high score desirable)	EQ-5D- VAS (high score desirable)	SSQ (Low score desirable)	MSPSS (high score desirable)	CSI (low score desirable)	ZCCS (low score desirable)
Residence	t (df 459) = .218, p=.827	**t (df 458) = -2.18; p=0.029 (Urban caregivers reported the lowest HRQoL)**	t (df 459) =1.44, p=.151	t (df 459) =-.002, p=.978	**t (df 459) =3.36; p< .001(Urban caregivers reported the greatest caregiver burden)**	t (df 459) =1.86, p=.064
Severity of CP	Current effect: F (4, 453) =1.9210, p=.106	Current effect: F (4, 452) =1.94, p=.103	Current effect: F (4, 453) =1.178, p=.320	Current effect: F (4, 453) =1.66, p=.157	**Current effect: F (4, 453) =8.6501, p<.001 (caregivers of more severely disabled children reported greater burden)**	**Current effect: F (4, 453) =12.090, p<.001 (caregivers of more severely disabled children reported greater burden)**

Child's age	**r= -0.134, p=0.017**	**r= -.137, p=.003**	**r=.236, p<.001**	**r= -.163, p<.001**	**r=.262, p<.001**	**r=.240, p<.001**
Duration of caregiving	**r= -0.136, p=.022**	**r= -.135, p=.004**	**r= 249, p< .001**	**r= -0.163, p=.001**	**r=261, p<.001**	**r=.238, p<.001**
Number of comorbid conditions	r= -.020, p=664	**r=-.113, p=.015**	**r=.098, p=.036**	**r=-.093, p=. 045**	**r=.170, p< .001**	**r=.184, p=.001**
Marital status	Current effect: F (3, 455) =1.80, p=.146	**Current effect: F (3, 454) =3.80, p=.010 (widows & divorced caregivers had the lowest HRQoL)**	Current effect: F (3, 455) =.421, p=.738	Current effect: F (3, 455) =1.92, p=.126	Current effect: F (3, 455) =1.99, p=.114	**Current effect: F (3, 455) =5.3133, p=.00132 (widows & divorced caregivers reported the highest caregiver burden)**
Educational status	**Current effect: F (3, 453) =4.51, p=.004 (more educated caregivers had the greatest HRQoL)**	**Current effect: F (3, 452) =6.04, p< .001 (more educated caregivers had the greatest HRQoL)**	**Current effect: F (3, 453) =9.86, p<. 001 (more educated caregivers had the least psychiatric morbidity)**	Current effect: F (3, 453) =2.39, p=.068	**Current effect: F (3, 453) =4.86, p=.00245 (more educated caregivers had the least caregiver burden)**	**Current effect: F (3, 453) =10.14, p=.00000 (more educated caregivers had the least caregiver burden)**
Employment status	Current effect: F (2, 457) =.271, p=.763	**Current effect: F (2, 456) =5.253, p=.006 (employed caregivers had greater HRQoL)**	Current effect: F (2, 457) =1.86, p=.157	**Current effect: F (2, 457) =3.55, p=.030 (employed caregivers had greater SS)**	**Current effect: F (2, 457) =4.35, p=.013 (unemployed reported the greatest burden)**	Current effect: F (2, 457) =1.13, p=.322
SES	**Current effect: F (4, 456) =15.80, p<.001 (Poorer caregivers reported the lowest HRQoL)**	**Current effect: F (4, 455) =17.380, p<.001 (Poorer caregivers reported the lowest HRQoL)**	**Current effect: F (4, 456) =20.155, p<.001 (Poorer caregivers reported the greatest psychiatric morbidity)**	**Current effect: F (4, 456) =7.1793, p<.001 (Poorer caregivers reported the least SS)**	**Current effect: F (4, 456) =20.465, p<.001 (Poorer caregivers reported the greatest caregiver burden)**	**Current effect: F (4, 456) =19.714, p<.001 (Poorer caregivers reported the greatest caregiver burden)**

N=431

8.11.6.2 Structural equation modelling

Illustrated in Figure 8-9 below is the final model denoting caregivers' mental health. The model explained 82.2% of the variance (

Table 8.17- Page 204). Apart from the likelihood ratio and normed chi-square, the model displayed fit against a set-criterion (Table 8.16).

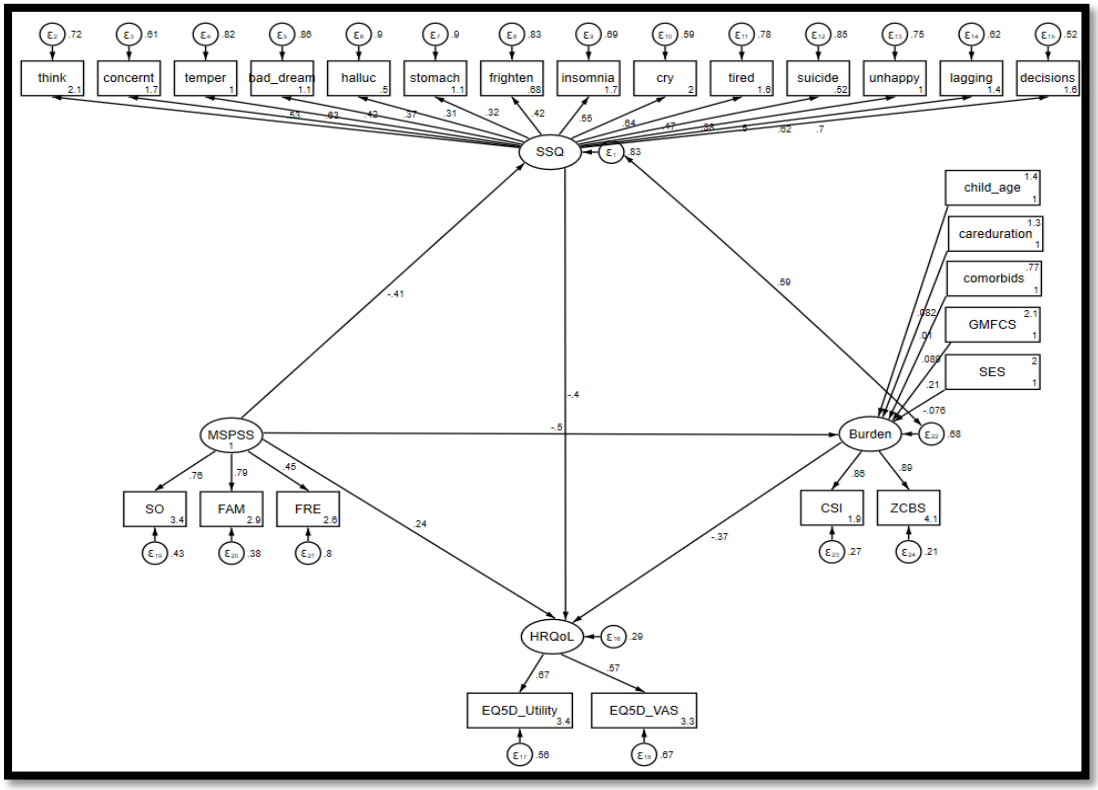


Figure 8-9: Model of the mental health of caregivers of children with CP

Table 8.16: Caregiver mental health model fit indices

Fit statistic	Index	Criterion for fit	Result – interpretation
Likelihood ratio	Chi-squared Test ($\chi ms2$)	p >0.05	χ2 (df 273) =2978.3 p<0.001- misfit
	Normed Chi-square [$\chi 2/df$]	$\chi 2/df$ <2	10.9- misfit
Population error	Root mean squared error of approximation (RMSEA)-(90% CI)	RMSEA≤ 0.06	0.053 (0.047: 0.058) -good fit
Information criteria	Akaike's information criterion (AIC)	Smaller = better	28936.7- best fit
	Bayesian information criterion (BIC)	Smaller = better	29233.7 – best fit
Baseline comparison	Comparative fit index (CFI)	CFI≥0.90	0.886 – optimal fit
	Tucker-Lewis index (LFI)	LFI≥0.90	0.905 – good fit
Size of residuals	Standardized root mean squared residual (SRMR)	SRMR≤ 0.08	0.082- good fit
	The coefficient of determination (SD)	Greater = better	0.882: optimal fit

Table 8.17: Total variance accounted for by the final SEM

		Variance explained					
		fitted	**predicted**	**residual**	**R-squared**	**mc**	**mc2**
Observed variables	think	0.151	0.042	0.109	0.278	0.527	0.278
	concernt	0.186	0.073	0.113	0.391	0.625	0.391
	temper	0.250	0.044	0.206	0.178	0.422	0.178
	bad_dream	0.248	0.034	0.215	0.135	0.368	0.135
	halluc	0.160	0.016	0.145	0.097	0.312	0.097
	stomach	0.247	0.025	0.222	0.100	0.317	0.100
	frighten	0.217	0.037	0.179	0.173	0.416	0.173
	insomnia	0.193	0.059	0.134	0.307	0.554	0.307
	cry	0.160	0.066	0.094	0.412	0.642	0.412
	tired	0.201	0.045	0.155	0.225	0.474	0.225
	suicide	0.169	0.025	0.145	0.146	0.382	0.146
	unhappy	0.250	0.062	0.188	0.248	0.498	0.248
	lagging	0.228	0.088	0.140	0.385	0.620	0.385
	decisions	0.204	0.099	0.105	0.484	0.696	0.484
	EQ5D_Utility	0.041	0.018	0.023	0.442	0.665	0.442
	EQ5D_VAS	352.135	115.351	236.785	0.328	0.572	0.328
	SO	1.139	0.651	0.488	0.572	0.756	0.572
	FAM	1.275	0.793	0.482	0.622	0.789	0.622
	FRE	1.297	0.263	1.034	0.203	0.451	0.203
	CSI	11.020	8.092	2.929	0.734	0.857	0.734
	ZCCS	544.496	430.248	114.247	0.790	0.889	0.790
Latent variables	SSQ	0.042	0.007	0.035	0.172	0.414	0.172
	HRQoL	0.018	0.013	0.005	0.709	0.842	0.709
	Burden	8.092	2.620	5.472	0.324	0.569	0.324
	Overall variance explained				**0.822**		

N=431

8.12 Discussion

This section of the study achieved the objectives set and both the psychometrics of the scales were established, and a profile of caregiver challenges emerged which confirmed that caring for a child with functional limitations does lead to an alteration in caregivers' mental health.

8.12.1 Methodological limitations

The disparity in some of the key demographics (child's age, place of residence, and dissimilar sample sizes) is a potential threat to the quality of the psychometric analysis, and the subsequent estimation of the model explaining the mental health of caregivers. Firstly, children with CP were significantly older than TDC. The discrepancies can be accounted for by the recruitment method, participants were conveniently selected. Further, most children presenting at either clinics, and vaccination sites were generally young. It would have been ideal if participants were matched for age as the burden of care may increase with the increased age of the child regardless of condition (CP vs. TDC). For example, the amount of physical burden (report of musculoskeletal pain) secondary to lifting, and transfers is likely to increase as the child grows older and heavier [9,29-31]. In the present study, rural-based participants were under-represented (13% vs. 87%), despite that 67% of the Zimbabwean population resides in rural areas [59]. Unfortunately, there are no exact statistics on the epidemiological distribution of CP cases against place of residence. We speculate that the prevalence of CP to be greater in urban areas because of better access to healthcare, and better epidemiological surveillance. A previous study has shown that the extrapolated prevalence of CP to be 1.55/1000 and 3.3/1000 in rural, and urban areas respectively [25]. The underrepresentation of CCWCP (461 vs. 771) could also have influence the study outcomes. Careful attention was made to adjust for the dissimilarity during analysis, for example, proportions/percentages instead of absolute frequencies were used in establishing association between categorical variables using Chi-Square tests.

8.12.2 Psychometric properties

The internal consistency of both the ZCCS and the MSPSS was high at sub-scale and scale levels, thus demonstrating reliability. Both instruments displayed evidence of construct validity and known-groups validity as caregivers of children with CP exhibited significantly increased caregiver challenges. This was true of the total score of all domains in the MCPSS and ZCCS apart from the physical and economic burden domain. Concurrent validity was also demonstrated in that there was a strong, positive correlation between scores on the ZCCS and CSI. In addition, caregivers with greater caregiver strain exhibited greater psychiatric morbidity and lower HRQoL and increased social support was associated with fewer caregiver challenges and higher HRQoL.

The final SEM model accounted for 82.2% of the variance which is further evidence of the construct validity of both instruments. The relationships between caregiver burden, SS, HRQoL and psychiatric morbidity were consistent with previous studies and caregiver burden conceptual frameworks [62,85,97,99,100]. The high correlation between CSI and ZCBS is a demonstration of the stability of

items on the ZCBS given that the CSI had been previously validated in Zimbabwean caregivers of children with CP [124].

8.12.3 Caregivers' mental health

As with previous studies and systematic reviews [83,493-495], the current study confirms that caring for a child with functional limitations leads to an alteration in caregivers' mental health. Current evidence also supports the multidimensionality of caregiver burden with financial strain, time pressure, change in personal plans and feeling overwhelmed by the caregiving role being cited as the greatest indicators of caregiver burden. Literature posits that caregiver burden is a product of cumulative stressors under the Double ABC-X model [107-109]. A stressor is defined as an occurrence which can potentially alter an individuals' well-being, for example, an inability to meet all caregiving expenses [105-109]. Results from univariate analysis concurred with previous studies as caregiver strain was exacerbated by the increased passage of time, the number of co-morbid conditions (a proxy indicator of the severity of CP), increase in child's age and being unemployed. For example, direct caregiving costs are likely to increase as the utilisation of medical services and specialist services is likely to increase as the child ages [136,494,508-511]. However, the addition of these contextual factors added "noise" to the final model and the model fit improved by the stepwise omission of some of the demographic variables. This again illustrates the complexity of conceptualization of caregiver burden as it is a context-specific latent variable [62,85,97,99,100].

Caregivers of children with CP reported the least amount of SS, and this is consistent with previous studies [136,137,141-146]. Due to the excessive demands of caregiving, carers are likely to have decreased opportunities for socialisation and engagement in hobbies and this negatively impacts their social network [138-145]. Results from univariate analysis further support this proposition as caregivers who were widows reported the least SS. Having a supportive spouse has been previously cited as a buffer to life stressors [145]. Further, consistent with the caregiver burden conceptual model by Raina et al. model [42], Double ABC-X model [107-109] and transactional model of psychological stress and coping [105-107], SS also evolved as a buffer to caregiver burden as it directly mediated the effects of caregiver burden and psychiatric morbidity and optimized caregivers' HRQoL. Caregivers who received an adequate amount of SS are likely to report a decrease in the CMDs such as anxiety and depression, improved HRQoL and decreased caregiver burden [29,48-50]. However, SS had a weaker direct effect on HRQoL. This suggests that there is a need to consider other interventions to improve caregiver mental health in addition to providing SS.

Caregivers received the least amount of SS from friends, similar to studies done in Malawi [47,563], Uganda [3], and Nigeria [54,555,564] which are countries of an almost similar socio-cultural context.

The discrepancies in sources of SS can be attributed to cultural influences whereby the immediate family (significant other and family) are invariably obliged to assist in the caregiving process, in which case the crisis (diagnosis of CP) is considered a shared burden within the family [27,145].

A greater amount of psychiatric morbidity was also associated with lower HRQoL and increased caregiver burden. The caring process is associated with physical, social and emotional distress [62,98,100]. For instance, anxiety, depression and hopelessness are very common in caregivers [13,16,48,52,218]. Unfortunately, poor mental health consequently leads to poor HRQoL and this may negatively affect the caregivers' capacity to provide quality care [603]. For instance, most children are reliant upon caregivers in the performance of basic ADLs such as bathing and feeding and routine rehabilitation such as execution of the prescribed home exercise programs and intake of medications [11,26]. Lack of resources [SS, financial and education] was also linked to poor HRQoL and greater psychiatric morbidity. Unfortunately, the prevalence and impact of CP are greater in people of lower SES, and poor caregivers are unlikely to have equitable access to coping resources. For example, poorer caregivers are unlikely to receive/access specialist support services, and this perpetuates a vicious circle of caregiver distress [52].

8.12.4 Conclusion

Collectively, the current evidence demonstrates the construct validity of the ZCCS and MSPSS and concurrent validity of the ZCCS. Higher caregiver burden was associated with greater psychiatric morbidity, and lower HRQoL and caregivers who received more of social support had the best mental health outcomes. Further, the final SEM also provided evidence of the multidimensionality of caregiver burden. Contextual factors, i.e. increased child age, increased caregiving duration, lower child functional capacity/more severe CP, and lower SES were associated with poorer mental health functioning.

9 Chapter 9: Conclusions and recommendations

This chapter gives an overview of the study outcomes, a critique of the study methodology and recommendations for practice and research.

9.1 Synthesis of study outcomes

The present study aimed to validate caregiver mental health outcomes and identify determinants of caregivers' mental health functioning within the context of Shona speaking Zimbabweans. This was attained through a three-fold process; firstly, we developed a context-specific caregiver burden outcome measure. The preliminary ZCCS item pool was developed through a synthesis of findings from a systematic review (Chapter Three), qualitative interviews with caregivers of children with CP (Chapter Four) and a content validation process by a panel of experts (Chapter Five). After that, the scale was validated on a large scale as respondents were recruited from both urban and rural areas. We then applied both CCT and IRT methods for item reduction; the resultant 33-items solution was multidimensional. Four factors were retained, i.e. physical and economic burden, concerns for the child, family relations and community participation. The ZCCS is a reliable tool as it yielded excellent IC ratings at both sub-scale [α range: .765-.841] and scale level [α=.925], and the ICC rating (95% CI) for ZCCS scores at baseline and after four weeks was 0.880 (0.793: 0.930) demonstrating the stability of the ZCCS.

In the second phase, the MSPSS was translated, adapted and validated into Shona, a Zimbabwean native language. The translation was preceded by a systematic review of the psychometric properties of translated versions of MSPSS. The SR aimed to identify translated tools, appraise their psychometrics and provide guidance in the development of a robust methodology for the translation of the MSPSS into Shona leveraging on a critique of existent translations. Based on the recommendations of the SR, a multi-stage translation process was utilised in translating the MSPSS, and caregivers' satisfaction with the translation was a testimony of a robust and accurate translation process. As with the ZCCS, the MSPSS was administered to a large pool of caregivers residing in both rural and urban areas. We applied both CTT and IRT methods to evaluate the dimensionality of the MSPSS. Both EFA and CFA replicated the original three-factor structure. However, the MSPSS-Shona did not meet all requirements for Rasch modelling, i.e. unidimensionality, local independence, and ordered thresholds. Dichotomisation of scoring options and the deletion of item 5 resulted in a parsimonious solution as the 11-items solution met Rasch modelling requirements. Both CTT and IRT methods provided evidence for the reliability of the MSPSS. The MSPSS-Shona version yielded excellent IC ratings at both sub-scale [α range: .833-.892] and scale level [α=.8685], and the ICC rating

(95% CI) for MSPSS scores at baseline and after four weeks was .980 (.959: 990) demonstrating the stability of the MSPSS. Further, the MSPSS yielded the following IRT reliability indices: person separation index (PSI) -0.731 and Cronbach alpha (α)- 0.722 and these imply perfect targeting and right scale reliability.

Findings from the third phase demonstrated the construct validity of the ZCCS and MSPSS and concurrent validity of the ZCCS. Higher caregiver burden was associated with greater psychiatric morbidity, and lower HRQoL and caregivers who received a higher amount of social support had the best mental health outcomes. Further, the final SEM also provided evidence of the multidimensionality of caregiver burden. Contextual factors, i.e. increased child age, increased caregiving duration, lower child functional capacity/more severe CP, and lower SES were associated with poor mental health functioning in caregivers of children with CP.

9.2 Critique of the study methodology

The study outcomes need to be interpreted with caution given some methodological limitations. Firstly, for the qualitative study, participants were recruited from only two institutions, and all participants were female. The institution-based recruitment of participants and solitary backward translations and coding of scripts might have reduced the study rigour and trustworthiness [487,523,528,604]. On the other hand, our study interview guide was based on well-established conceptual frameworks [42,106-109], and themes which evolved in the qualitative study were "replicated" during factor analysis, univariate analysis and SEM. The concruency with quantitative outcomes is suggestive of the internal and external validity of the qualitative study. Above all, to the best of our knowledge, this is a first study to qualitatively investigate the experiences of Zimbabwean caregivers of children with CP. This study can act as a template for further exploration of caregivers' experiences.

The ZCCS was developed through the amalgamation of qualitative input from caregivers, synthesis of a systematic review, content validation by a panel of experts and caregivers' cognitive briefing interviews. Further, the utilisation of both traditional (factor analysis) and modern (Rasch analysis) psychometric methods for item reduction is a strength of the study. The exclusion of only 12 of the initial 45-item bank is a further testimony of the robustness of the item development and content validation process. However, the ZCCS did not meet all the requirements of Rasch modelling, and thus it should be treated as an ordinal scale. This is a potential weakness as the use of an ordinal scale may lead to inaccurate conclusions when performing parametric tests such as SEM [170,180-186]. However, ZCCS data were normally distributed, and the use of large sample size can offset the potential limitations. There is, therefore, need for further studies to refine the metric properties of

the ZCCS in line with the increased propensity towards the use of modern psychometric methods [191,192,194,196,197]. Our earlier SR indicated that most caregiver burden outcome measures used in low resourced settings were generic and were not adequately adapted and validated. The ZCCS is on the other end of the spectrum as it is a context-specific outcome measure with visible evidence of reliability and validity.

The designing of the methodology for the translation of the MSPSS was based on a previous systematic review, and this increased the internal and external validity of our study based on the COSMIN criterion. Further, use of innovative methods like making use of pictorial prompts to enhance participants' understanding of the scoring instructions [3,47,563] also increased the methodological rigour as testified by the replication of the three-factor solution using both EFA and CFA. However, we had to drop an item and dichotomise responses to attain an 11-item, parsimonious solution during Rasch modelling. The discrepancies between CTT and Rasch model solutions are defensible given the methodological and philosophical differences between the two psychometric methods [168,194]. Further studies are needed to compare the performance of the 12- and 11-items solutions. Nevertheless, this is the first study which has used both CTT (EFA and CFA) and IRT (Rasch analysis) in evaluating the psychometrics of the MSPSS. Above all, the MSPSS exhibited evidence of reliability, stability, construct validity and known-groups validity.

Children with CP were significantly older than TDC, and the age discrepancy is a threat to both the internal and external validity of the study. The demands of caregiving increase with an increase in child age, for instance, children become heavier as they grow, and this may increase the prevalence of musculoskeletal disorders such as shoulder and low back pain in caregivers [124]. Ideally, both sets of children should have been matched for age, and this is a limitation of the study. Further, the study selection criterion was based on self-report data, and this could have negatively influenced the study outcomes. For example, it was important to differentiate between existent chronic morbidities, and impairments secondary to caregiving. Methodologically, this was not possible as the two may share the same causal pathway, and this could be another potential methodological flaw. It would have been ideal to exclude caregivers with clinically confirmed diagnosis of an existent chronic condition(s) before the assumption of the caregiving role.

9.3 Recommendations

9.3.1 Future studies

Future qualitative studies need to explore the concepts of caregiver burden and social support. Emphasis should be placed on recruiting participants from multiple sites, recruiting community-dwelling caregivers, recruitment of male caregivers, triangulation of qualitative findings by multiple

designs such as focus group discussions and participant observation amongst other methods. There is a need for independent transcription and coding, member checks and keeping of audit trails to increase the rigour and trustworthiness of the qualitative studies [487,537,604].

There is also a need for longitudinal, quantitative studies to further understand the impact of caring for a child with a disability. The current political and socio-economic environment in Zimbabwe may have led to an "exaggeration" of caregiving difficulties/challenges. More so, there is a need to explore the positive benefits of caregiving [281,605,606]. The COSMIN guidelines explicitly state that the construct to be measured needs to be succinctly defined when developing a new PROM [148,150,595]. We thus explored only one perspective of caregivers' experiences, i.e. caregiver burden/challenges.

Future studies should focus on using modern psychometric methods such as Rasch analysis to develop true-metric and shorter-versions of the ZCCS to increase the research and clinical utility of the ZCCS. In its present state, the 33-items version may not be easily integrated for routine clinical use.

9.3.2 Physiotherapy practice

Given the requirement of evidence-based practice, there is a need for physiotherapists to integrate PROMs in routine care. This is especially important in low-resource settings such as Zimbabwe where there is a dearth of published information on the role of physiotherapy in mental health. Although physiotherapists are playing a crucial role in the management of long-term paediatric conditions such as CP, there is a need for local therapists to embrace locally-validated PROMS such as the ZCCS, MSPSS-Shona, SSQ, CSI and EQ-5D which were further validated using a large sample in the present study. Efforts should be made, however, to disseminate current evidence to clinicians using seminars in the hope of stimulating the need for the use of PROMs for both routine clinical practice and research purposes. Findings can also be used to inform the development of context-specific interventions for improving the mental health of caregivers. For instance, in addition to improving child outcomes, there is a need to provide care to caregivers who are at risk of common mental disorders. Physiotherapists can use the validated tools to screen the at-risk caregivers, offer psychosocial support and refer for specialist treatment where appropriate.

9.3.3 Policymakers

There is a need for policymakers to provide support to caregivers, for example, social grants will lead to improved caregivers' well-being. Economic burden evolved as a significant predictor to poor caregiver strain, so there is also need for the government to consider establishing micro-financing schemes to economically empower caregivers who are likely to have limited opportunities for income generation given the demands of the caregiving process and the associated expenses. Additionally,

there is a need to prioritise the funding of mental health programs such as caregiver well-being programs as a higher proportion of health financing is currently skewed towards endemic conditions such as TB, HIV/AIDS.

9.4 Conclusion

The current study contributes to the body of knowledge through the validation of caregivers' mental health outcomes and the determination of mental health functioning of caregivers residing in low-resource settings. The ZCSS and MPCSS can now be used with confidence within the context of Shona speaking caregivers and efforts should be made to integrate these PROMs into routine clinical care.

The challenges facing the caregivers of children with CP are considerable. It is hoped that the development and validation of appropriate tools to monitor the impact of caring will lead to improved awareness of the needs of the caregivers and their children. This, in turn, may lead to the development, implementation and evaluation of tailor-made caregiver wellness programs.

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11 Appendices

11.1 Appendix 1: Description of study participants and settings – Paediatric caregiver burden outcome measures systematic review

Name of tool	Authors [Year of publication]	Country - Income bracket	Design	Participants	Outcome measure-Domain(s)	Outcome measure-Domain(s)	Outcome measure-Domain(s)	Outcome measure-Domain(s)	Outcome measure-Domain(s)
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Bemister et al. [2015]	Canada- HIC	Longitudinal	Parents of children with perinatal stroke, N=103	The Hospital Anxiety and Depression Scale-Anxiety & depression	The Perceived Stress Scale (PSS) - perceived stress	The Pediatric Quality of Life Inventory-Family Impact Module (PedsQL) - Quality of life and family functioning	The Dyadic Adjustment Scale (DAS)- Marital strain	
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Bemister et al. [2014a]	Canada- HIC	Longitudinal	Families of children with perinatal stroke, N=110	The Hospital Anxiety and Depression Scale-Anxiety & depression	The Perceived Stress Scale (PSS) - perceived stress	The Pediatric Quality of Life Inventory-Family Impact Module (PedsQL) - Quality of life and family functioning		
Alberta Perinatal Stroke Project Parental Outcome Measure (APSP-POM)	Bemister et al. [2014b]	Canada- HIC	Longitudinal	1. Mothers of children with perinatal stroke, n=86 2. Mothers of typically developing children, n=62	The Hospital Anxiety and Depression Scale-Anxiety & depression	The Perceived Stress Scale (PSS) - perceived stress	The Pediatric Quality of Life Inventory-Family Impact Module (PedsQL) - Quality of life and family functioning	The Dyadic Adjustment Scale (DAS)- Marital strain	
Assessment of Caregiver	Matsumoto et al. [2011]	USA-HIC	Longitudinal	Caregivers of children with	Gross Motor Function	The Pediatric Evaluation of	Pediatric Outcomes Data Collection		

Experience with Neuromuscular Disease (ACEND)				neuromuscular disorders, N=60	Classification System (GMFCS) - the severity of CP	Disability Inventory-functioning	Instrument - functioning		
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)	Difazio et al. [2016]	USA-HIC	Longitudinal	Caregivers of children with CP, N=43	Caregiver Priorities and Child Health Index of Life with Disabilities (CPCHILD)- children with CP HRQoL	Gross Motor Function Classification System (GMFCS) - the severity of CP			
Assessment of Caregiver Experience with Neuromuscular Disease (ACEND)	Vessey et al. [2017]	USA-HIC	Prospective observational	Caregivers of children with CP, N=52	Family Expense Diary	Impact on Family Scale- caregiver strain on family			
Autism Parenting Stress Index (APSI)	Silva & Schalock [2011]	USA-HIC	Cross-sectional	1. Autism, n=107 2. Typically developing children, n=139 3. Developmental disabilities, n=28					
Autism Parenting Stress Index (APSI)	Silva et al. [2011]	USA-HIC	Random controlled trial	Parents of children with Autism, N=42	Autism Behaviour Checklist- autistic behaviour	Pervasive Developmental Disorders Behaviour Checklist -behaviour & language	Vineland Adaptive Behavior Scales, 2nd Edition - behaviour	Autism Behavior Checklist (ABC)- behaviour	Sense and Self-Regulation Checklist (SSC)- sensation
Autism Parenting Stress Index (APSI)	Silva et al. [2015]	USA-HIC	Random controlled trial	Parents of children with Autism, N=84	Childhood Autism Rating Scale, 2nd Edition, Standard - the severity of autism	Preschool Language Scale, 5th Edition (PLS-5)- language			
Caregiver Burden Index (CBI)	Liu et al. [2015]	Taiwan-HIC	Mixed methods	Caregivers of children with allergies, N=124	None				

Caregiver Burden Index (CBI)	Ekim et al. [2017]	Turkey-UMIC	Cross-sectional	Caregivers of children with allergies, N=213	None			
Caregiver Burden Inventory (CBIn)	Rani et al. [2013]	Pakistan-LMIC	Cross-sectional	Caregivers of children with CP, N=196	Caregiver stress self-assessment scale - stress levels			
Caregiver Burden Scale (CBS)	Cavalari et al [2017]	Brazil-UMIC	Cross-sectional	Caregivers of children myelomeningocele, N=26	Pediatric Evaluation of Disability Inventory - functioning			
Caregiver Burden Scale (CBS)	Rubira et al [2012]	Brazil-UMIC	Cross-sectional	Caregivers of children with cancer, N=160	SF-36- HRQoL			
Caregiver Burden Scale (CBS)	Piran et al. [2017]	Iran - UMIC	Cross-sectional	Caregivers of children with chronic illnesses, N=249	Ad-hoc- clinical and demographics of children			
Caregiver Burden Scale (CBS)	Santo et al [2011]	Brazil-UMIC	Cross-sectional	Caregivers of children with cancer, N=32	SF-36- HRQoL	Beck's Depression Inventory-Depression		
Caregiver Difficulties Scale (CDS)	Wijesinghe et al. [2013]	Sri Lanka	Mixed methods	Cerebral palsy, N=190	WHOQOL-BREF - HRQoL	The severity of CP - Physician evaluation		
Caregiver Difficulties Scale (CDS)	Wijesinghe et al [2015]	Sri Lanka	Cross-sectional	Cerebral palsy, N=375	Adhoc questionnaire			
Caregiver Difficulties Scale (CDS)	Farajzadeh et al [2018]	Iran - UMIC	Cross-sectional	Caregivers of children with CP, N=216	Caregiver burden scale - caregiver burden	Beck depression inventory (BDI-II)-depression	Fatigue severity scale- fatigue	WHOQOL-BREF - QOL
Caregiver Strain Index (CSI)	Jubber et al [2013]	USA-HIC	Cross-sectional	Type-1 Diabetes, N=85	Psychological Control Scale questionnaire -	HbA1c values - metabolic control	Porter-O'Leary Scale - marital conflict	

					Psychological control				
Caregiver Strain Index (CSI)	Dambi & Jelsma [2014]	Zimbabwe-LIC	Quasi-experimental	Cerebral palsy, N=46	EQ-5D - HRQoL	GMFM-66 - Motor functioning	GMFCS- Severity of CP		
Caregiver Strain Index (CSI)	Dambi et al [2015]	Zimbabwe-LIC	Longitudinal	Caregivers of children with CP, N=46	EQ-5D - HRQoL	GMFM-66 - Motor functioning	GMFCS- Severity of CP		
Caregiver Strain Index (CSI)	Luescher et al. [199]	USA	Cross-sectional	Caregivers of children with Joubert Syndrome, N=49	Beck Depression Inventory - depression	Child development inventory - illness severity	Ways of Coping Questionnaire-Revised - coping strategies		
Caregiver Strain Index (CSI)	Dyches et al. [2016]	USA-HIC	Cross-sectional	Single mothers of children with autism spectrum disorder, N=122	Ad-hoc questionnaire				
Caregiver Strain Index (CSI)	Tsai & Wang [2008]	Taiwan-HIC	Cross-sectional	Mothers of intellectually disabled children, N=127	Social support scale-social support				
Caregiver Strain Questionnaire (CSQ)	Iadarola et al. [2017]	USA-HIC	Random controlled trial	Parents of children with Autism, N=180	Parenting Stress Index-Short Form - stress	Parenting Sense of Competence (PSOC) -parental satisfaction and efficacy	Aberrant Behavior Checklist (ABC) - child's behaviour	The Autism Diagnostic Observation Schedule (ADOS)- behaviour observation	Mullen Scales of Early Learning (MSEL) - cognitive functioning
Caregiver Strain Questionnaire (CSQ)	Kirby et al [2015]	USA-HIC	Cross-sectional	Parents of children with neurodevelopmental disabilities, N= 97	Sensory Experiences Questionnaire (SEQ) - sensory features	Mullen Scales of Early Learning (MSEL) - cognitive functioning	Stanford-Binet Intelligence Scales (SB5) - intelligence		
Family Burden Interview Schedule (FIBS)	Suresh et al. [2014]	India- LMIC	Prospective	Parents of children with neurodevelopmental disabilities, N= 97	The Sensory Processing Measure-sensory processing impairment	Brief Autism Mealtime Behaviour Inventory (BAMBI) - feeding problems	Childhood Autism Rating Scale- the severity of autism	Gesell's Developmental Schedule- diagnosis of intellectual disability	The Vineland Social Maturity Scale- social skills

Family Burden Interview Schedule (FIBS)	Khanna et al. [2015]	India- LMIC	Cross-sectional	Caregivers of children with chronic illness, N=204	Patient Health Questionnaire (PHQ-9)- psychological wellbeing	Generalized Anxiety-- psychological wellbeing Disorder (GAD-7)			
Family Impact of Childhood Disability (FICD)- original version	Trute et al. [2007]	Canada-HIC	Longitudinal	Parents of children with developmental disabilities, n=158.	The Rosenberg Self-Esteem scale - self-esteem	Family Assessment Measure III - Brief Form (FAMBF) - family functioning			
Family Impact of Childhood Disability (FICD)- original version	Ooh & Magiati [2014]	Singapore-HIC	Cross-sectional	Families of children with autism spectrum disorders, N=65	The Questionnaire on Resources and Stress – Friedrich Short Form (QRS-F)- family functioning	The Coping Health Inventory for Parents (CHIP) - coping strategies	The Family Adaptability and Cohesion Evaluation Scales IV (FACES IV) – family adaptation		
Family Impact of Childhood Disability (FICD)- original version	Schlebusch et al. [2016]	South Africa-MIC	Cross-sectional	Families of children with autism spectrum disorders, N=180	The Family Routines Inventory (FRI)- family routines	Beach Center Family Quality of Life Scale (FQOL) - the family quality of life			
Family Impact of Childhood Disability (FICD)- original version	Trute et al. [2002]	Canada-HIC	Longitudinal	Parents of children with developmental disabilities, n=158.	Parenting Stress Index short form (PSI)- parental stress	Dyadic Adjustment Scale (DAS) marital adjustment	Family Adjustment Measurement III - Short Form (FAM-IIISF) - family adjustment	Family Coping Strategies Scale (F-COPES) - family coping	Beck Depression Inventory (BDI) - depression
Family Impact of Childhood Disability (FICD)- original version	Guyard et al. [2012]	European countries - HICs	Cross-sectional	Parents of children with CP, N=242	McMaster Family Assessment Device (GF)- family functioning	Parenting Stress Index short form (PSI)- parental stress			
Family Impact of Childhood Disability (FICD)- original version	Thompson et al. [2007]	Canada- HIC	Longitudinal	Parents of children with developmental disabilities, n=158.	The Rosenberg Self-Esteem scale - self-esteem	Family Assessment Measure III - Brief Form (FAMBF) - family functioning	Parenting Stress Index short form (PSI)- parental stress		

Family Impact of Childhood Disability (FICD)- revised version	Guyard et al [2017]	Ireland, France & Denmark - HICs	Cross-sectional	Parents of children with CP, N=220	Parenting Stress Index short form (PSI)- parental stress	Family Assessment Device Agreement scale- family functioning	Bimanual Fine Motor Function Classification (BFMF) -manual skills	Gross Motor Function Classification System (GMFCS)- motor function	
Family Impact Questionnaire (FIQ)	Ingersoll et al. [2016]	USA-HIC	Random controlled trial	Parents of children with Autism, N=27	Parent Sense of Competence Scale (PSOC) - competence	MacArthur-Bates Communicative Development Inventory (MCDI)- child's expressive vocabulary	Vineland Adaptive Behavior Scales ((VABS-II)-child adaptive functioning		
Family Impact Questionnaire (FIQ)	Boström et al. [2010]	Sweden - HIC	Cross-sectional	Parents of children with intellectual disability, n=216	EASI Temperament Survey- child temperament				
Family Impact Questionnaire (FIQ)	Neece [2014]	USA-HIC	Quasi-experimental	Parents of young children with developmental delays, N=46	Parenting Stress Index-Short Form (PSI-SF) - parenting stress	Center for Epidemiologic Studies Depression Scale (CES-D) - depression screening	Satisfaction with Life Scale (SWLS)- satisfaction with life	The Child behaviour checklist (CBCL)- child behaviour	Subjective Units of Distress Scale (SUDS)- parenting distress
Family Impact Questionnaire (FIQ)	Neece [2012]	USA-HIC	Cross-sectional	Parents of children with developmental delays y, n=93 2. Parents of typically developing children, n=144	Stanford-Binet IV (SB-IV - children's cognitive ability	Child Behaviour Checklist (CBCL) - child's behaviour			
Family Impact Questionnaire (FIQ)	Eisenhower et al. [2005]	USA-HIC	Cross-sectional	Parents of young children with intellectual disability, N=215	Bayley Scales of Infant Development The BSID-II - child development	Child Behaviour Checklist (CBCL) - child's behaviour	Stanford-Binet IV (SB-IV - children's cognitive ability	Center for Epidemiologic Studies Depression Scale (CES-D) - depression	
Family Impact Questionnaire (FIQ)	Milshtein et al. [2010]	Israel- HIC	Cross-sectional	Parents of young children with Autism, N=61	The WAIS-IIIHEB - parents intelligence	Autism-Spectrum Quotient (AQ)- autistic traits	Autism Diagnostic Interview-Revised (ADI-R)- diagnostic tool	Autism Diagnostic Observation Schedule (ADOS)- a diagnostic tool	Vineland Adaptive Behavior Scales

									(VABS)- behaviour
Family Impact Questionnaire (FIQ)	Olsson & Hwang [2008]	Sweden - HIC	Cross-sectional	1.Parents of children with intellectual disability, n=111 2. Parents of typically developing children, n=319	Beck Depression Inventory - depression	Ad-hoc questionnaire - risk factors			
Fathers of Children with Developmental Challenges Questionnaire- (FCDC)	Ly & Goldberg [2014]	USA-HIC	Cross-sectional	Fathers of children with developmental delays, N=85	Parenting Stress Index (PSI)- parental stress	Parenting Commitment Scale (PCS) - parenting commitment	NEO Five-Factor Inventory (NEO-FFI) - personality assessment	Social Communication Questionnaire (SCQ) - communication behaviour	
Impact on Family Scale (IPFAM) - original version	Antiel et al. [2016]	USA-HIC	Random controlled trial	Caregivers of children with spina bifida, N=175	Parenting Stress Index short form (PSI)- parental stress	Family Support Scale (FSS) - social support	Family Resource Scale (FRS)- family resources		
Impact on Family Scale (IPFAM) - original version	Ryan et al. [2009]	Canada-HIC	Quasi-experimental	Caregivers of children with CP, N=29	Family Impact of Assistive Technology Scale (FIATS)				
Impact on Family Scale (IPFAM) - original version	Şimşek et al [2014]	Turkey-UMIC	Cross-sectional	Caregivers of children with physical disabilities, N=352	WeeFIM- child functioning				
Impact on Family Scale (IPFAM) - original version	Carlson & Miller [2017]	USA-HIC	Cross-sectional	Caregivers of children with physical epilepsy, 1=352	Interpersonal Support Evaluation List (ISEL)- social support	COPE - stress and coping	Questionnaire on Resources and Stress – Fredrich edition (QRS-F)		
Impact on Family Scale (IPFAM) - original version	Stuart & McGrew [2009]	USA-HIC	Cross-sectional	Caregivers of children with Autism, N=78	Gilliam Autism Rating Scale, Second Edition (GARS-2) severity of autism	Social Readjustment Rating Scale (SRRS) - stressful events	Multidimensional Scale of Perceived Social Support (MSPSS) - social support	Family Implications of Childhood Disability Scale (FICD) - family distress; Dyadic	Brief COPE - coping mechanisms; caregiver strain

								Adjustment Scale (DAS) -marital burden	questionnaire - caregiver burden
Impact on Family Scale (IPFAM) - original version	Fonsea et al [2015]	Portugal-HIC	Cross-sectional	Parents of infants with congenital abnormalities, N= 79	Kansas Inventory of Parental Perceptions (PCS) - parenting perceptions	Parenting Stress Index short form (PSI)- parental stress			
Impact on Family Scale (IPFAM) - original version	Hsieh et al. [2014]	Taiwan-HIC	Correlational	Parents of Children with developmental delays, N=60	Pediatric Outcomes Data Collection Instrument (POCDI)- child functioning	Child Health Questionnaire – Parental Form 28 (CHQ-PF28) – child HRQoL	WHOQOL-BREF-QoL	Hospital Anxiety and Depression Scale (HADS) - anxiety & depression	
Impact on Family Scale (IPFAM) - original version	Hutchinson et al [2009]	USA-HIC	Cross-sectional	Caregivers of children with brain tumours, N=90	Brief Symptom Inventory [BSI] - psychological morbidity	Impact of Event Scale [IES] - traumatic events	Parent Experience of Child Illness [PECI]- parental adjustment	Caregiver Strain Questionnaire [CGSQ]- caregiver burden	
Impact on Family Scale (IPFAM) - original version	Heath et al [2006]	Australia - HIC	Cross-sectional	Caregivers of children with cancer, N=56	Finances diary- expenditure				
Impact on Family Scale (IPFAM) - original version	Hsieh et al. [2013]	Taiwan-HIC	Cross-sectional	Caregivers of children with developmental disability, N=70	PedsQL-Family Impact Module	WHOQOL-BREF-QoL	PedsQL-health satisfaction questionnaire		
Impact on Family Scale (IPFAM) - original version	Wright et al. [2005]	Canada-HIC	Quasi-experimental	Caregivers of children with CP, N=9	Gross Motor Function Measure (GMFM)- functioning	Quality of Upper Extremity Skills Test (QUEST)- dexterity	Peabody Developmental Motor Scales- functioning	Paediatric Evaluation of Disability Inventory (PEDI)- functioning	Pictorial Scale of Perceived Competence and Social Acceptance for Young Children
Impact on Family Scale	Majnemer et al. [2012]	Canada- HIC	Cross-sectional	Caregivers of children with CP, N=87	Vineland Adaptive Behavior Scale	Gross Motor Function Measure	Child Health Questionnaire (CHQ)- health status	Parenting Stress Index short form	

(IPFAM) - original version					(VABS)- child behaviour	(GMFM)- functioning		(PSI)- parental stress
Impact on Family Scale (IPFAM) - original version	Blacher et al. [2013]	USA-HIC	Cross-sectional	Parents of Children with Intellectual Disabilities, N=112	Bayley Scales of Infant Development II (BSID-II)- development			
Impact on Family Scale (IPFAM) - original version	Seliner [2016]	Switzerland-HIC	Cross-sectional	Parents of Children with profound disabilities, N=117	SF-36- HRQoL	Measurement of Process of Care (MPOC-20)- satisfaction with care	DISABKIDS Smiley- Child HRQoL	
Impact on Family Scale (IPFAM) - revised version	Stein & Jessop [2003]	USA-HIC	Cross-sectional	Parents of children with chronic conditions, N=630	The child Functional Status (FS)- functioning	The Personal Adjustment and Role Skills Scale (PARS)- child psychological adjustment	Psychiatric Symptom Index- maternal psychiatric morbidity	
Impact on Family Scale (IPFAM) - revised version	Kao et al. [2009]	USA-HIC	Cross-sectional	Families of children with chronic illness and developmental disability, N=122	Brief Symptom Inventory (BSI) - psychological symptoms	Child Behavior Checklist (CBCL) - behaviour		
Impact on Family Scale (IPFAM) - revised version	Brown et al. [2012]	Canada-HIC	Cross-sectional	Parents of children with autism, N=97	Family Needs Questionnaire (FNQ) - family needs	Scales of Independent Behaviour-Revised (Short Form)- child/s behaviour		
Maslach Burnout Inventory (MBI)	Karadavut & Uneri [2011]	Turkey-UMIC	Cross-sectional	Mothers of infants with brachial plexus injury, N=18	Beck Depression Inventory - depression			
Maslach Burnout Inventory (MBI)	Esma et al [2011]	Turkey-UMIC	Cross-sectional	Caregivers of children with CP, N=115	Gross Motor Function Classification System (GMFCS) - severity of CP			

Maslach Burnout Inventory (MBI)	Basaran et al. [2013]	Turkey-UMIC	Cross-sectional	1. Caregivers of children with CP, n=143 2. Caregivers of typically developing children, n=60	Beck Depression Inventory - depression	WHOQOL-BREF-QOL	Gross Motor Function Classification System (GMFCS) - the severity of CP
Maslach Burnout Inventory (MBI)	Bilgin & Gozum [2009]	Turkey-UMIC	Random controlled trial	Mothers with an intellectually disabled child, n=90	Ad-hoc- clinical and demographics of children		
Modified Caregiver Strain Index (M-CSI)	Chiluba & Moyo [2017]	Zambia-LMIC	Mixed methods	Caregivers of children with CP, N=25	The Rehabilitation and Support Questionnaire-caregivers' services perception		
Modified Caregiver Strain Index (M-CSI)	Sharan et al. [2012]	India- LMIC	Cross-sectional	1. Caregivers of children with CP, n=257 2. Caregivers of ambulatory children with other orthopaedic problems, n=117	Borg CR-10 scale-physical exertion		
Modified Caregiver Strain Index (M-CSI)	Panganiban-Corales [2011]	Philippines - LMIC	Cross-sectional	Caregivers of children with cancer, N=90	Filipino Family APGAR- coping strategies	SCREEM Family Resources Survey (SCREEM-RES)-family coping strategies	
Nijmeegse Ouderlijke Stress Index (NOSI)	van der Veek et al. [2009]	Netherlands-HIC	Cross-sectional	Caregivers of children with Down Syndrome, N=621	Cognitive Emotion Regulation Questionnaire (CERQ)- cognitive coping strategies		
Parental Stress Scale (PSS)	Huang et al. [2014]	Taiwan-HIC	Cross-sectional	1.fathers of children with developmental disabilities, n=206 2.	Short form questionnaire (SF-36)-HRQoL	The Chinese version of parental stress scale- stress	

				fathers of healthy children, n=207					
Parental Stress Scale (PSS)	Norizan & Shamsuddin [2010]	Malaysia-HIC	Cross-sectional	Mothers of children with Down syndrome, N=147	Depression, Anxiety and Stress Scale 21-item -DASS	COPE Inventory	Paediatric Symptom Checklist (PSC)		
Parental Stress Scale (PSS)	Tellegen & Sanders [2014]	Australia-HIC	Random controlled trial	Parents of children with ASD, N=64	The Parenting Tasks Checklist -PTC	Depression, Anxiety and Stress Scale 21-item -DASS	Family Observation Schedule -FOS	Parent Problem Checklist -PPC	Relationship Quality Index - RQI 2. The Goal Achievement Scales (GAS)
Parents of Children with Disabilities Inventory (PCDI)	Noojin & Wallander [1996]	USA-HIC	Cross-sectional	Mothers of children with CP and spina bifida, N=111	Severity of Physical Handicap Scale (SHS)- child's disability	Brief Symptoms Inventory - mothers' mental health	Wahler Physical Symptoms Inventory (WPSI) - physical health	Questionnaire on Resources and Stress-Short Form (QRS-SF)- stress	Family Adaptability and Cohesion Evaluation Scales III (FACES III)- dissatisfaction with family functioning
Parents of Children with Disabilities Inventory (PCDI)	Gloria et al. [2016]	Hong Kong - HIC	Cross-sectional	Caregivers of children with disabilities, N=131	Barthel Index - functional level	Affiliate Stigma Scale - stigma	Mental Health Inventory-18 - anxiety and depression	Multidimensional Scale of Perceived Social Support- social support	
Perceived Stress Scale -10	Lightsey & Sweeney [2008]	USA-HIC	Cross-sectional	Mothers of children with disabilities, n=64	The coping inventory for stressful situations-coping	The generalised self-efficacy scale - GSE	The meaning of life questionnaire	The family satisfaction scale - FSS	The family environment scale third edition - FES-3
Perceived stress scale-10	Lovell et al. [2016]	UK-HIC	Quasi-experimental	Caregivers of children with autism, n=33	Strength and Difficulties Questionnaire (SDQ) - problematic child behaviours	The Hospital Anxiety and Depression Scale (HADS)- Anxiety and depression			
Perceived stress scale-10	Tehee et al. [2008]	Ireland-HIC	Cross-sectional	Parents of children with ASD, N =42	The Involvement and responsibility questionnaire-	The family stress and coping questionnaire(FSCQ-	The Support questionnaire (SQ)-	Information and education questionnaire (IEQ)-	

					parental responsibility	A) -stress and coping	Helpfulness of support systems	information on ASDs	
Perceived stress scale-10	Giallo & Gavidia-Payne [2006]	Australia-HIC	Cross-sectional	1.Siblings of children with disability, n=49 2. Parents of children with disability, n=49 3. Children with a disability, n=49	The Siblings Daily Hassles stress and uplifts scale-stress	The Self report coping scale- coping	The strengths and difficulties questionnaire Parent Version-Emotional and behavioural functioning of children	The Parent Behaviour Questionnaire-parent's use of parenting skills	The Family; Hardiness, problem-solving communication and family time and routines indices
Perceived stress scale-10	Huang et al. [2014]	Taiwan-HIC	Cross-sectional	1.Fathers of children with developmental disabilities, n=206 2. Fathers of healthy children, n=207	Short form questionnaire (SF-36)-HRQoL				
Perceived stress scale-10	Barlow et al. [2008]	UK- HIC	Randomized control trial	Parents of children with disabilities, N=188	Generalized Self-Efficacy Scale (GSES)	Parent's Self-Efficacy Scale	Hospital Anxiety and Depression Scale (HADS)	Satisfaction with Life Scale (SWLS)	Visual Analogue Scale (VAS)
Perceived stress scale-10	Lovell et al. [2015]	UK- HIC	Correlation	Caregivers of children with 1. Autism N=14 2. ADHD N=4	Hospital Anxiety and Depression Scale (HADS)	Pennebaker Inventory of Limbic Languidness (PILL)-physical health	Strength and Difficulties Questionnaire (SDQ)	Cortisol levels - stress	
Perceived stress scale-10	Lovell et al. [2012]	UK- HIC	Cross-sectional	1. Caregivers of children with Autism and ADHD N=67	Interpersonal support evaluation checklist	Levels of Cortisol	Plasma levels of proinflammatory biomarkers		
Perceived stress scale-14	Mak & Ho [2007]	China-HIC	Cross-sectional	Caregivers of children with intellectual disability, N=212	Modified family support scale -social support	COPE Inventory-coping strategies	Relationship focused coping scale-coping strategies	Caregiver burden inventory-negative perceptions	Kansas inventory of parental perceptions-positive perceptions
Perceived stress scale-14	Lightsey & Sweeney [2008]	USA-HIC	Cross-sectional	Mothers of children with disabilities, n=64	The coping inventory for stressful situations-coping	The generalised self-efficacy scale(GSE)-generalised self-efficacy	The meaning of life questionnaire-the presence and search for meaning	The family satisfaction scale(FSS)- overall satisfaction with	The family environment scale third edition(FES-3)-family, social and

								family adaptability and cohesion	environmental characteristics
Perceived stress scale-14	Chen et al. [2015]	Chile-HIC	Cross-sectional	Caregivers of children with disabilities, N=90	smoking status	saliva telomere length			
Perceived stress scale-14	Benn et al. [2012]	USA-HIC	Random controlled trial	1. Parents, n=25 2. Educators, n=35	The Five facet mindfulness questionnaire-mindfulness	The State trait anxiety inventory (STAI) for adults-anxiety	The centre of epidemiological studies depression scale(CES-D)-Depression	The positive and negative affect schedule(PANAS)-Positive and negative affect	The psychological wellbeing scale-personal growth; The parenting stress index
Perceived stress scale-14	Gallagher et al. [2009]	UK- HIC	Longitudinal	1. Parents of children with developmental disabilities, n=30 2. Parents of typically developing children, n=29	Venous blood sample- antibody titre	The Hospital anxiety and depression scale(HADS)-depression	The support functions scale-social support	The caregiver burden index-caregiver burden	The strength and difficulties questionnaire-child problem behaviour; PSQI
Perceived stress scale-14	Skok et al. [2006]	Australia-HIC	Cross-sectional	Mothers of children with CP, N=43	The Gross motor function classification system (GMFCS)-severity of disability	The Multidimensional scale of perceived social support (MSPSS)- social support	The Profile of adaptation to life (Clinical scale) (PAL-C)- Psychological adjustment, functioning and physical health	The satisfaction with life scale(SWLS)-Life satisfaction	The psychological well-being subscale of PAL-C and SWLS- well being
Perceived stress scale-14	Al-Gamal et al. [2013]	Jordan-MIC	Cross-sectional	Parents of children with cerebral palsy, n=204	The Gross motor function classification system (GMFCS)-severity of disability	The Perceived Stress Scale (PSS) - perceived stress	The Beck depression inventory-depression	The strengths and difficulties questionnaire-psychological well being	The Multidimensional scale of perceived social support (MSPSS)-social support
Perceived stress scale-14	Mclean et al. [2015]	Australia-HIC	Cross-sectional	Parents of children with obstetrical	The Parents of children with disabilities inventory (PCDI)-	The General health questionnaire-12(GHQ-12)-			

				brachial plexus injuries, N=52	disability related stress	psychological distress			
Perceived stress scale-14	Findler [2014]	Israel- HIC	Cross-sectional	1.Grandparents of children with intellectual disability, n=94 2. Grandparents of children without intellectual disability, n=105	The Multidimensional experience of grandparenthood set of inventories(MEG)-grand parenthood experiences	The Multidimensional scale of perceived social support (MSPSS)-social support	The level of differentiation of self-scale(LDSS)-emotional functioning	The family adaptability and cohesion evaluation scale (FACES-III)-family functioning	Posttraumatic Growth Inventory (PTGI)-psychological functioning
Perceived stress scale-4	Cantwell et al. [2014]	Ireland-HIC	Cross-sectional	Parents caring for children with developmental disabilities, N=167	Personal mastery scale (PMS) - self-mastery	Support functions scale (SFS) - social support	Strengths and difficulties questionnaire (SDQ)-child's behaviour	Physical Health Questionnaire (PHQ) - physical health	
Perceived stress scale-4	McConnell et al. [2015]	Canada-HIC	Cross-sectional	Families of children with disabilities, N=538	Developmental behaviour checklist (DBC-24)- child's behaviour	Oslo-3 Social Support Scale (OSS-3)- social support	Placement Tendency Index (PTI)-placement propensity		
Perceived stress scale-4	McConnell et al. [2016]	Canada-HIC	Longitudinal	Families of children with disabilities, N=538	Developmental behaviour checklist (DBC-24)- child's behaviour	Oslo-3 Social Support Scale (OSS-3)- social support	Placement Tendency Index (PTI)-placement propensity		
Perceived stress scale-4	Stephen Gallagher & Whiteley [2013]	UK- HIC	Cross-sectional	1.Parents of children with intellectual disabilities, n=70 2. Parents of typically developing children, n=45	Physical Health Questionnaire (PHQ) - physical health	Support functions scale (SFS) - social support	Strengths and difficulties questionnaire (SDQ)-child's behaviour		
PSI- Long Form (PSI-LF)	Gupta [2007]	USA-HIC	Cross-sectional	Parents of children with 1. ADHD n=50 2. Developmental delays n=28 3.HIV or Asthma n=46 4. Control n=22	Adhoc-demographic questionnaire				

PSI- Long Form (PSI-LF)	Gulsrud et al. [2016]	USA-HIC	Randomised controlled trial	Parents of children with ASD, N=86	Videotaped Parent-child interaction	Mullen Scales of Early Learning (MSEL)	Reynell Developmental Language Scales	Caregiver Involvement Scale	
PSI- Long Form (PSI-LF)	Keller & Honig [2004]	USA-HIC	Cross-sectional	Parents of elementary-school-aged children requiring special education services N=30	Family Environment Scale (FES)	Family Support Scale			
PSI- Long Form (PSI-LF)	Brahm et al. [2016]	Australia-HIC	Longitudinal	Parents of children with ASD, N= 152	Parenting Alliance Measure (PAM)	Autism Specific Parenting Self-Efficacy (ASPSE)			
PSI- Long Form (PSI-LF)	Gong et al. [2015]	China-HIC	Cross-sectional	1.Parents of children with autism spectrum disorders n=188 2. Control n=144	Autism Behavior Checklist (ABC)	Childhood Autism Rating Scale (CARS)	Self-Rating Depression Scale (SDS)	Self-Rating Anxiety Scale (SAS)	
PSI- Long Form (PSI-LF)	Vermaes et al. [2008]	Netherlands-HIC	Cross-sectional	Parents of children with spina bifida, N=83	Physical Dysfunction	Wechsler Intelligence Scale for Children (WISC-III)	Quick Big Five (QBF)		
PSI- Long Form (PSI-LF)	Guralnick et al. [2006]	USA-HIC	Longitudinal	Caregivers of children with mild developmental delays N=63	Wechsler Preschool and Primary Scale of Intelligence-revised (WPPSI-R)	Wechsler Intelligence Scale for Children (WISC-III)	Test for Auditory Comprehension of Language-Revised (TACL-R)	Expressive One-Word Picture Vocabulary Test-Revised (EOWPVT-R)	Vineland Adaptive Behaviour Scales 2. Inventory of Parental Experiences (IPE)
PSI- Long Form (PSI-LF)	Delambo et al. [2011]	USA-HIC	Cross-sectional	Parents of children with developmental disability, N=48	Adhoc-demographic questionnaire				
PSI- Long Form (PSI-LF)	Wanamaker & Glenwick [1998]	USA-HIC	Cross-sectional	Parents of pre-schoolers with Cerebral Palsy, N=84	Parenting Sense of Competence Scale (PSOC)	Social Support Questionnaire-6 (SSQ-6)	Eyberg Child Behavior Inventory (ECBI)	Beck Depression Inventory (BDI)	

PSI-Short Form (PSI-SF)	Webster et al. [2008]	Australia-HIC	Longitudinal	Caregivers of children with developmental delay, N=65	VABS-child behaviour	child health questionnaire-health status	Battle Developmental Inventory-child development	
PSI-Short Form (PSI-SF)	Lin et al. [2011]	Taiwan-HIC	Random controlled trial	Caregivers of children with CP, N=22	Peabody Developmental Motor Scales II (PDMS-2)-hand use	Bruininks–Oseretsky Test of Motor Proficiency (BOTMP)- hand use	Caregiver Functional Use Survey (CFUS) - hand function	
PSI-Short Form (PSI-SF)	Ferre et al. [2015]	USA-HIC	Quasi-experimental	Caregivers of children with CP, N=15	daily logs	Assisting Hand Function	COPM	
PSI-Short Form (PSI-SF)	Kleefman et al. [2014]	Netherlands	Random controlled trial	Caregivers of children with intellectual disability, N= 111	Alabama parenting questionnaire-parenting	Eyberg child behaviour inventory - behaviour	SDQ- behaviour	
PSI-Short Form (PSI-SF)	Britner et al. [2003]	USA-HIC	Cross-sectional	1. Caregivers of children with CP, n=57 2. Caregivers of TDC, n=30	Family support scale-social support	Support functions scale-social support	Dyadic adjustment scale- family adjustment	
PSI-Short Form (PSI-SF)	Samadi et al. [2014]	Iran-UMIC	Cross-sectional	1. Parents who had a child with ASD, n=121 2. Parents of children with intellectual disabilities, n=115	The General health questionnaire-psychiatric morbidity	The McMaster family assessment device-family functioning	Satisfaction with caring	
PSI-Short Form (PSI-SF)	Meppelder et al. [2015]	Netherlands-HIC	Cross-sectional	Parents with children with Din=134	The Dutch version of the caregiver-teacher report form (C-TRF)-child behaviour problems	Teacher report form-child behaviour problems	The Dutch version of the Vineland adaptive behaviour scales- adaptive functioning	The Support interview guide-support network size
PSI-Short Form (PSI-SF)	Parkes et al. [2011]	*European countries-HICs	Cross-sectional	Parents with children with CP, N=818	The Gross motor function classification	The Bimanual fine motor function-use of arms and hands	IQ Assessment-intellectual capacity	The Child health questionnaire parent form- child bodily pain

					system-gross motor function				
PSI-Short Form (PSI-SF)	Theule et al. [2011]	Canada-HIC	Cross-sectional	Families with children with ADHD, n=95	Conners' rating scales-revised long version(CRS)-assess ADHD symptoms in children and adolescents	Conners' adult ADHD rating scales(CAARS)-assess parental ADHD symptoms	The Family support scale- social support	Wechsler abbreviated scale of intelligence(WASI)-intelligence	Wechsler intelligence scale for children, fourth edition (WISC-IV)-intelligence
PSI-Short Form (PSI-SF)	Minnes et al. [2015]	Canada-HIC	Cross-sectional	Mothers of children with disabilities, N=155	The Scales of independent behaviour revised early development form (SIB-R)child's adaptive behaviour	The Brief COPE-parent coping	The Family empowerment scale- parental self-efficacy	The Positive gain scale(PGS)- positive caregiving experiences	
PSI-Short Form (PSI-SF)	Kanaheswari et al. [2011]	Malaysia-HIC	Cross-sectional	Mothers of children with spinal bifida, N=81	The Vineland adaptive behaviour scales (VABS)-adaptive behaviour				
PSI-Short Form (PSI-SF)	Dardas & Ahmad [2014]	Jordan-UMIC	Cross-sectional	Parents of children with autistic disorder, n=184	The Ways of coping checklist revised - coping strategies used by individuals to deal with specific stressful events	The World health organisation Quality of life brief version-parents' quality of life			
PSI-Short Form (PSI-SF)	Oelofsen & Richardson [2006]	UK-HIC	Cross-sectional	Families of 1. preschool children with developmental disabilities, n=59 2. TDC n=45	The Orientation of life questionnaire - a sense of coherence	The Health perceptions questionnaire-health status	The Family support scale- social support		
PSI-Short Form (PSI-SF)	Sarimsk et al. [2013]	Germany-HIC	Longitudinal study	Caregivers of children with disabilities, N=125	The Family impact scale-family related stress	The General self-efficacy Germany Questionnaire-general self-efficacy	The Early intervention parenting self-efficacy scale (EIPSES)-parenting competence		

PSI-Short Form (PSI-SF)	Hassall et al. [2005]	UK-HIC	Cross-sectional	Mothers of children with intellectual disability, n=46	The Vineland adaptive behaviour scales-adaptive behaviours	The Family support scale-social support	The Parenting sense of competence scale-parenting self-esteem	The Parental locus of control scale short form (PLOC)-parenting control	The Vineland Maladaptive behaviour domain-behavioural difficulties
PSI-Short Form (PSI-SF)	Ong et al. [2011]	Malaysia-HIC	Cross-sectional	1. Parents of children with spina bifida(SB), n =66 2. Parents of non-disabled children, n=66	The General health questionnaire (GHQ-12)-mental health	The Vineland adaptive behaviour scale (VABS)-adaptive skills			
PSI-Short Form (PSI-SF)	Woolfson & Grant [2006]	UK-HIC	Cross-sectional	1. Parents of children with developmental disabilities(DD), n =53 2. Parents of typically developing (TD) children, n=60	Child-rearing practices report (CRPR) -parenting behaviour				
PSI-Short Form (PSI-SF)	Zaidman-Zait et al. [2014]	Canada-HIC	Longitudinal study	Mothers of children with ASD, n=184	The Child behaviour checklist for ages from 1 1/2 to 5-behavioural problems	The Vineland adaptive behaviour scale (VABS)-adaptive skills	The Preschool language scale (PLS)-receptive and expressive language	The Merrill-Palmer reversed scales of development-cognitive development	
PSI-Short Form (PSI-SF)	Hung et al. [2004]	China-HIC	Cross-sectional	1.Parents of disabled children, n= 92 2. Parents of children with cancer, n= 89	Demographic-Adhoc questionnaire				
PSI-Short Form (PSI-SF)	Huang et al. [2014]	Taiwan-HIC	cross-sectional	1. Caregivers of children with autism, n=52	The Childhood autism rating scale (CARS)- autistic behaviours	The Strength and difficulties questionnaire-Chinese version (SDQ-C) -emotional problems, behavioural			

						problems and strength			
PSI-Short Form (PSI-SF)	Tomanik et al. [2004]	USA-HIC	Cross-sectional	Mothers of children with a pervasive developmental disorder, n=60	The Aberrant behaviour checklist-child aberrant behaviour	AAMR Adaptive behaviour scales-child adaptive behaviour			
PSI-Short Form (PSI-SF)	Brei et al. [2015]	USA-HIC	Cross-sectional	1.Parents of children with ASD, n=19 2. Parents of children without ASD, n=21	The Early learning composite score from the Mullen scales of early learning- cognitive ability	The Vineland adaptive behaviour scales (VABS)-adaptive behaviour	The Aberrant behaviour checklist-behavioural problems	The Child behaviour checklist (CBCL)-behavioural problems	The Autism diagnostic observation schedule (ADOS)-autism severity
PSI-Short Form (PSI-SF)	Macias et al. [2007]	USA-HIC	Cross-sectional	Two-parent families of a child with spina bifida, n= 71	The Family resource scale (FRS)-parental perceived adequacy of resources	The Family support scale (FSS)-social support	Kaufman Brief intelligence test (K-BIT)- child cognitive development	The Child behaviour checklist parent report (CBCL)-maladaptive behaviour	The Social skills rating system (SSRS)- social skills
PSI-Short Form (PSI-SF)	Zaidman-Zait et al [2010]	Canada-HIC	Cross-sectional	Parents of children with ASD, n =141					
PSI-Short Form (PSI-SF)	Smith et al. [2015]	Canada-HIC	Quasi-experimental	Children with ASD, n=118	Preschool language scale fourth edition (PLS-4) - language	The Merrill Palmer revised scales of development(M-P-R)- cognition	The Vineland adaptive behaviour scale (VABS)-adaptive skills	The Social responsiveness scale (SRS)-autism symptom severity	The Child behaviour checklist (CBCL)-child behaviour problems
PSI-Short Form (PSI-SF)	Kieffer-Kristensen et al. [2013]	Denmark-HIC	Cross-sectional	Families in which a parent has acquired a brain injury, n= 35	Children's revised impact of event scale (CRIES)- assess post-traumatic stress disorder	The Child behaviour checklist-emotional and behavioural problems	The Dyadic adjustment scale (DAS)- marital satisfaction	The Symptom checklist- 90 - Revised (SCL-90-R)-depression, anxiety and aggression	The European brain injury questionnaire (EBIQ)- brain injury symptoms
PSI-Short Form (PSI-SF)	Foody et al. [2015]	Ireland-HIC	Cross-sectional	1.Parents of children with ASD, N=38	The Hospital anxiety and depression	The Parental responsibility scale	Salimetrics oral swabs- salivary collection	Oscar ABP monitor-cardiovascular assessment	

					scale(HADS)- anxiety and depression	(PRS)- parental responsibility			
PSI-Short Form (PSI-SF)	Georgiades et al. [2011]	Canada-HIC	Longitudinal study	1.Caregivers of children with ASD, N=335	The Autism diagnostic interview-revised (ADI-R)	The Child behaviour checklist - behavioural problems	The Repetitive behaviour scale-revised (RBS-R)- behaviour	The Vineland adaptive behavioural scales second edition (VABS II)- child adaptive behaviour	The Merrill-Palmer reversed scales of development-intellectual ability
PSI-Short Form (PSI-SF)	Stadnick et al. [2015]	USA-HIC	Quasi-experimental	1. parent of children with ASD, N=30	The Social communication questionnaire (SCQ)- ASD symptoms	The Social responsiveness scale (SRS)- severity of autistic symptoms	The Modified checklist for autism in toddlers (M-CHAT)- ASD screening for children 16-30 months	The Vineland adaptive behaviour scales second edition (Vineland-II)-adaptive functioning	The Center for epidemiological studies depression scale (CES-D)-depression
PSI-Short Form (PSI-SF)	Dykens et al. [2014]	USA	Randomised controlled trial	Mothers of children with disability, N=243	The Beck depression inventory (BDI)-depression	The Beck anxiety inventory (BAI)-anxiety	The Insomnia severity index- sleep insomnia	The Ryff scales of psychological well-being short form-psychological well being	The Life satisfaction scale-life satisfaction
PSI-Short Form (PSI-SF)	Solomon et al. [2008]	USA-HIC	Quasi-experimental	Parents of children with ASD, N= 19	The Eyberg child behaviour inventory-disruptive child behaviour problems	The Behaviour assessment system for children Parent Rating Scales (BASC)- behaviour and emotion	The Shared Positive Affect coding - Parent-child shared effect		
PSI-Short Form (PSI-SF)	Giovagnoli et al. [2015]	Italy-HIC	Quasi-experimental	1.Parents of children with ASD, n=190 2. Parents of typically developing children, n= 122	The Autism diagnostic observation schedule-generic (ADOS-G) - autistic symptoms	The Child behaviour checklist (CBCL) - emotional and behavioural problems			
PSI-Short Form (PSI-SF)	Brossard-Racine et al. [2012]	Canada-HIC	Cross-sectional	1.Parents of children with CP, N=76	The Strengths and difficulties questionnaire (SDQ)- behaviour	The Gross motor function classification	Leiter Intelligence test -cognitive ability	The Vineland adaptive behavioural scales Interview edition	

						system-gross motor function		(VABS)- social and personal functioning	
PSI-Short Form (PSI-SF)	Warfield et al. [2014]	USA-HIC	Cross-sectional	1. Caregivers of children with ASD, N=74	Gittelle's relational coordination measure-coordination between formal and informal providers				
PSI-Short Form (PSI-SF)	Hoffman et al. [2009]	USA-HIC	Quasi-experimental	1.Mothers of children with autism, n= 104 2. Mothers of TDC, n= 342	The Gilliam autism rating scale second edition (GARS-2) - evaluates autism symptoms				
PSI-Short Form (PSI-SF)	Feizi et al. [2014]	Iran-UMIC	Cross-sectional	Mothers of children with disabilities, N=285	Demographic-Adhoc questionnaire				
PSI-Short Form (PSI-SF)	Bagner & Eyberg [2007]	USA-HIC	Randomised controlled trial	Families of children with mental retardation, N=30	The Wechsler preschool and primary scale of intelligence - third edition (WPPSI-III) - cognitive ability	The Adaptive behaviour scale-school: Second edition (ABS-S:2)-adaptive behaviour	The Childhood autism rating scale (CARS) -identifies children with autism and distinguishes them from developmentally disabled children without autism	The Wonderlic personnel test (WPT) -adult intellectual abilities	The Diagnostic interview schedule for children - fourth edition -Parent version (DISC-IV-P) - mental disorders
PSI-Short Form (PSI-SF)	Davis & Carter [2008]	USA-HIC	Longitudinal study	Parents of children with ASD, N=108	The Beck anxiety inventory (BAI) - anxiety	The Center for epidemiologic studies depression inventory(CES-D)-depression	The Autism diagnostic observation schedule-generic (ADOS-G)- assesses social and communicative functioning in individuals suspected to have ASD	The Autism diagnostic interview-revised(ADI-R)-diagnosis of autism	The Mullen scales of early learning -overall developmental composite score

PSI-Short Form (PSI-SF)	Hodge et al. [2013]	USA-HIC	Cross-sectional	1.Mother-child dyad of children with ASD, N=180	The Gilliam autism rating scale (GARS-2)- the severity of ASD symptoms	The Children's sleep habits questionnaire (CSHQ)- sleep quality	Pittsburgh sleep quality index (PSQI)- maternal sleep	The Symptom assessment-45 Questionnaire(SA-45)- maternal mental health	
PSI-Short Form (PSI-SF)	Majnemer et al. [2008]	Canada-HIC	Cross-sectional	Families of children with CP, N=95 B	The Pediatric quality of life inventory (PedsQL) -quality of life	The Dimensions of mastery questionnaire- mastery motivation 2. The Strengths & difficulties questionnaire	The Vineland adaptive behaviour scale (VABS)- adaptive skills	The Children's assessment of participation and enjoyment (CAPE) - participation and enjoyment	Leiter intelligence test- intelligence; The impact on family scale (IOF)
PSI-Short Form (PSI-SF)	Ketelaar et al. [2008]	Netherlands-HIC	Cross-sectional	Parents of children with CP, N= 42	The Vineland adaptive behaviour scales (VABS)- adaptive behaviour	The Paediatric evaluation of disability inventory (PEDI)- functional skills			
PSI-Short Form (PSI-SF)	Most et al. [2006]	USA-HIC	Longitudinal	1. mothers of children with down syndrome, n= 25 2. Mothers of young children with mixed aetiology, n =49	The Bayley scales of infant development (BSID-II) - development	The Differential ability scales (DAS)- cognitive ability	The MacArthur communicative development inventory (CDI) - child's lexical development	The Infant temperament questionnaire (ITQ)- child's temperament	The Child behaviour checklist (CBCL)- child behaviour problems
PSI-Short Form (PSI-SF)	Al-Khalaf et al. [2014]	Jordan-UMIC	Quasi-experimental	1.Parents of preschool children with ASD, N=20	The Coping strategy indicator (CSI)- situational coping				
PSI-Short Form (PSI-SF)	Braiden et al. [2012]	UK-HIC	Quasi-experimental	Parents of children with ASD, N=31	The Psycho-educational profile 3- assessment of skills and behaviours of children with autism				
PSI-Short Form (PSI-SF)	Tervo [2010]	USA-HIC	Cross-sectional	Caregivers of children with ASD, N=281	The Child development inventory-	The Child behaviour checklist- behavioural problems	The Inventory for client and agency planning- behaviours		

					developmental assessment				
PSI-Short Form (PSI-SF)	Bagner & Graziano [2013]	USA-HIC	Cross-sectional	Parents of children with developmental delay N=44	The Wonderlic personnel test-maternal cognition	The Wechsler abbreviated scale of intelligence-intelligence	The Wechsler preschool and primary scale of intelligence third edition (WPPSI)-cognitive ability	The Bayley scales of infant and toddler development third edition -cognitive ability	The Child behaviour checklist (CBCL)-child behaviour problems; The Parental distress scale
PSI-Short Form (PSI-SF)	Jeter et al. [2017]	USA-HIC	Cross-sectional	Primary caregivers of children with ASD, N=335	The Eyberg child behaviour inventory-disruptive child behaviour problems	The Behavioural assessment system for children second edition (BASC-2)-child functioning			
PSI-Short Form (PSI-SF)	Tervo [2012]	USA-HIC	Cross-sectional	Caregivers of children with global delay, N=201	The Child development inventory-developmental assessment	The Child behaviour checklist - behavioural problems	The Diagnostic and statistical manual oriented scale-emotional problems		
PSI-Short Form (PSI-SF)	Samadi et al. [2013]	UK-HIC	Quasi-experimental	Parents of children with ASD, N=37	The General health questionnaire-wellbeing	The Coping styles questionnaire(CSQ)-coping strategies	The Family functioning scale-family functioning		
PSI-Short Form (PSI-SF)	Minjarez et al. [2013]	USA-HIC	Quasi-experimental	Families of children with autism, N=17	The Family empowerment scale(FES)- family, service and community empowerment				
PSI-Short Form (PSI-SF)	Kurtz-Nelson & McIntyre [2017]	USA-HIC	Cross-sectional	Parents of children with developmental delay, N=119	The Child behaviour checklist - child behaviour problems	The Revised life orientation test (LOT-R)-optimism	The Parent feelings questionnaire(PFQ)-positive and negative feelings		
PSI-Short Form (PSI-SF)	Benzies et al. [2013]	Canada-HIC	Cross-sectional	Mothers of children with disability, N=154	The Brief Family assessment	The General self-efficacy scale			

					measure III(FAM)- family functioning	(GSES)- personal competence			
PSI-Short Form (PSI-SF)	Bannink et al. [2016]	Uganda-LIC	Cross-sectional	Parents of children with spina bifida N=134	The Vineland adaptive behaviour scales-adaptive behaviours	The Daily functioning subscales- child's daily functioning level			
PSI-Short Form (PSI-SF)	Valicenti-Mcdermott et al. [2015]	USA-HIC	Cross-sectional	1. Families of children with autism, n=50 2. Families of children with other developmental disabilities(DD), n=50	The Gastrointestinal questionnaire-gastrointestinal and feeding problems	The Child sleep habits questionnaire- sleep disturbances	The Aberrant behaviour checklist - behaviour		
PSI-Short Form (PSI-SF)	Craig et al. [2016]	Italy-HIC	Cross-sectional	1.Parents of children with neurodevelopmental disorders, n=239 2. Parents of TDC, n=53	The Child behaviour checklist(CBCL) - behaviour problems	The Leiter international performance scale revised-Intelligence	The Wechsler intelligence scale for children (WISC-III)-intelligence	The Wechsler preschool and primary scale of intelligence(WPPSI)-intelligence	
PSI-Short Form (PSI-SF)	Bennett et al. [2013]	UK-HIC	cross-sectional	Parents of children with brain tumours, n=37	The Parental locus of control scale (PLOC)- parental appraisals	Ways of coping questionnaire (Revised)- parenting coping styles			
PSI-Short Form (PSI-SF)	Padden & James [2017]	UK-HIC	Cross-sectional	1.Parents of children with ASD, n=38 2. Parents of TDC, n=38	The Gilliam autism rating scale (GARS-2)- assesses severity of ASD symptoms	The Hospital anxiety and depression scale (HADS)-anxiety and depression	The Parental responsibility scale (PRS)- parenting responsibility	The Brief COPE-coping strategies 2. The Pittsburgh sleep quality index (PSQI)	The Social support questionnaire Short form (SSQ)-social support
PSI-Short Form (PSI-SF)	Hill-Chapman et al. [2013]	USA-HIC	Cross-sectional	Parents of children with autism spectrum disorders N=56	Behaviour Assessment System for Children (BASC-2)	Parenting Alliance Inventory (PAI)			

PSI-Short Form (PSI-SF)	Benzies et al. [2009]	Canada-HIC	Cross-sectional	Canadian mothers of children with disabilities N=195	Parenting Morale Index (PMI)	Family Impact of Childhood Disability (FICD)	Brief Family Assessment Measure (FAM)	Personal Well-Being Index 2. General Self-Efficacy Scale (GSES) 3. S	Positive and Negative Affect Schedule
PSI-Short Form (PSI-SF)	Ello & Donovan [2005	USA-HIC	Cross-sectional	Parents of children with developmental disability N=64	Adhoc-demographic questionnaire				
PSI-Short Form (PSI-SF)	Rivard et al. [2014]	Canada-HIC	Cross-sectional	Families of children with autism spectrum disorders, N= 236	Childhood Autism Rating Scale (CARS)	Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III)	Adaptive Behaviour Assessment System-II (ABAS-II)	Parenting Stress Index-Short Form	
PSI-Short Form (PSI-SF)	Moss et al. [2014]	USA-HIC	Quasi-experimental	Families with children with developmental disabilities N=26	Developmental Behaviour Checklist: Parent Version	Caregiver Acceptance of Treatment Survey	Goal Attainment Scale (GAS)		
PSI-Short Form (PSI-SF)	Smith et al. [2014]	USA-HIC	Cross-sectional	Parents of children with 1. Down's Syndrome N=29 2. Other developmental disabilities N=82	Parent Perception of Language Development (PPOLD)	Sequenced Inventory of Communication Development-Revised (SICD-R)	Vineland Adaptive Behaviour Scales		
PSI-Short Form (PSI-SF)	Parkes et al. [2009]	Ireland-HIC	Cross-sectional	Caregivers of children with CP, N=102	Child Health Questionnaire (CHQ) - well-being	Strengths and Difficulties Questionnaire (SDQ)- behaviour			
PSI-Short Form (PSI-SF)	Ahmad & Dardas [2015]	USA-HIC	Cross-sectional	Fathers of children with ASD, N=101	The World Health Organization Quality of Life Assessment (WHOQOL-BREF)				
PSI-Short Form (PSI-SF)	Dunn et al. [2012]	USA-HIC	Quasi-experimental	Parents of children with ASD, n=20	The Sensory profile-child's sensory experiences	The Parenting sense of competence scale-parenting self-efficacy	The Canadian occupational performance measure(COPM)-child participation	The Goal attainment scaling (GAS)-goal progress in everyday life	

PSI-Short Form (PSI-SF)	Trute et al. [2005]	Canada-HIC	Longitudinal	Parents of children with disabilities N=151	Family Needs Survey (FNS)	Family Assessment Measure-Brief Form (FAM-BF)			
PSI-Short Form (PSI-SF)	Darling et al. [2011]	USA-HIC	Cross-sectional	1.Fathers of children with disabilities, n=85 2. Fathers of children without disabilities, n=121	The Family inventory of life events (FILE)	The Parenting daily hassles scale(PDHS)-daily stresses	The Family crisis oriented personal evaluation scale (FCOPES)	The Family health status inventory (FHSI)- health stress	The Satisfaction with life scale (SWLS)-life satisfaction
PSI-Short Form (PSI-SF)	Im-Bolter et al. [2015]	Canada-HIC	Cross-sectional	1. Mothers of children with mental health problems, N=50	The Blishen socioeconomic index for occupation in Canada-socioeconomic status	The Child behaviour checklist (CBCL)	Wechsler intelligence scales for children third edition (WISC-III)-verbal, performance and IQ score	The Clinical evaluation of language fundamentals-third edition (CELF-3)	
PSI-Short Form (PSI-SF)	Niccols & Mohamed [2000]	Canada-HIC	Quasi-experimental	Parents of children with developmental delay, N=17	The Parenting sense of competence scale- parenting competence	The Center for epidemiologic studies depression scale- parental depression	The Family assessment device general functioning scale- family functioning	Client satisfaction questionnaire-satisfaction	
PSI-Short Form (PSI-SF)	Dardas& Ahmad [2014]	Jordan-UMIC	Cross-sectional	Parents of children with autistic disorder, n=184					
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Cho & Hong [2013]	Korea- HIC	Cross-sectional	Caregivers of children with developmental disability, N=160	Beach Center Family Quality of Life Scale-QOL	Social support scale (SSS)-social support			
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Rickards et al. [2007]	Australia-HIC	Random controlled trial	Parents of children with autism and developmental delay, N=59	Vineland Adaptive Behavior Scale (VABS)- child behaviour	Mental Development Index (MDI)- cognition	Bayley Behaviour Rating Scale-behaviour		
Questionnaire on Resources and Stress-	Thomas et al. [2007]	USA-HIC	Cross-sectional	Caregivers of children with Autism, N=353	Ad-hoc- clinical and demographics of children				

Short Form (QRS-SF)									
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Reed et al. [2017]	UK- HIC	Cross-sectional	Caregivers of children with Autism, N=93	Social Communication Questionnaire (SCQ) - socialization	Strengths and difficulties questionnaire (SDQ)- child's behaviour			
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Vijesh et al. [2007]	India- LMIC	Cross-sectional	Caregivers of children with CP, N=50	Ad-hoc- clinical and demographics of children				
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Siman-Tov & Kaniel [2011]	Israel- HIC	Cross-sectional	Caregivers of children with Autism, N=176	Sense of Coherence Scale (SOC)- coherence	Locus of control scale (LCS)	Family Support Scale (FSS)- family support	Mental health scale (MHS)	Quality of marriage scale (QMS) ; Autism Behaviour Checklist (ABC)
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Wang et al. [2011]	China- HIC	Cross-sectional	Families with children with autism and other developmental disabilities, N=368	COPE Inventory				
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Küçüker [2006]	Turkey-UMIC	Cross-sectional	Parents of children with developmental disabilities, N=57	Beck Depression Inventory (BDI)				
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Reed & Osborne [2013]	UK- HIC	Cross-sectional	Caregivers of children with Autism, N=52	Gilliam Autism Rating Scale (GARS) - behaviour	psycho-educational Profile—Revised (PEP-R)- capabilities	Conners' Rating Scale (CRS-R)- behavioural problems		
Questionnaire on Resources and Stress-	Weinhouse et al. [1992]	USA-HIC	Cross-sectional	Parents of children with developmental disabilities, N=32	Ad-hoc- clinical and demographics of children				

Short Form (QRS-SF)									
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Shin & Crittenden [2003]	Korea & USA- HICs	Cross-sectional	Mothers of children with mental retardation, N=78	Traditional Values Scale (TVS)	Maladaptive Behavior Scale (MBS)	Multidimensional Scale of Perceived Social Support (MSPSS) - social support	Parental Attitude Research Instrument (PARI)	
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Honey et al. [2005]	UK- HIC	Cross-sectional	Caregivers of children with Autism, N=174	Judson Scale (JS)-parental adaptation	Family Support Scale (FSS)	Family Crisis Orientated Personal Evaluation Scales (F–COPES	Autism Behavior Checklist (ABC)	
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Watson et al. [2013]	Canada-HIC	Cross-sectional	Caregivers of children with Autism & fetal alcohol distress, N=50	Parenting Stress Index-Short Form (PSI-SF)				
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Lam et al. [2003]	UK- HIC	Cross-sectional	Caregivers of children with learning disabilities, N=47	Ways of Coping Questionnaire (WCQ)- coping mechanisms	Family Support Scale (FSS)- family support	General Health Questionnaire (GHQ) - physical health		
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Kaniel & Siman-Tov [2011]	Israel- HIC	Cross-sectional	Caregivers of children with Autism, N=166	Sense of coherence scale (SOC)-coherence	Locus of control scale (LCS)	Family Support Scale (FSS)- family support	Folkman questionnaire-threats	Mental health scale (MHS)
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Hastings et [2005]	UK- HIC	Cross-sectional	Caregivers of children with Autism, N=135	Hospital Anxiety and Depression Scale (HADS) - anxiety & depression	COPE inventory-coping			
Questionnaire on Resources and Stress-	Wong et al. [2006]	Hong Kong - HIC	Quasi-experimental	Caregivers of children with	Maternal Self-Rating Scale-efficacy	Community Activity Questionnaire (CAQ)	Parent Experience Survey (PES)-parental experiences		

Short Form (QRS-SF)				developmental disability, N=40					
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Stoneman [2007]	USA-HIC	Quasi-experimental	Caregivers of children with Down Syndrome, N=100	Center for Epidemiologic Studies Depression Scale (CES-D)-depression	Family Support Scale (FSS)- family support	Parental Attitudes towards Childrearing Questionnaire (PACQ)- attitudes	Temperament Assessment Battery (TAB)- child temperament	
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Saloviita et al. [2003]	Finland-HIC	Cross-sectional	Caregivers of a child with intellectual disability, N=236	Marital Adjustment Test (MAT)-family resources	Personal Assessment of Intimacy in Relationships (PAIR) – spousal support	Family Support Scale (FSS)- family support	Ways of Coping Checklist (Revised)-coping	Social Readjustment Rating - life meaning
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Hastings et al. [2002]	UK- HIC	Cross-sectional	Parents of children with Down's syndrome, N=61	Demographic Questionnaire				
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Lloyd & Hastings [2009]	UK- HIC	Cross-sectional	Caregivers of a child with intellectual disability, N=196	Hospital Anxiety and Depression Scale (HADS) - anxiety & depression	Trait Hope Scale (THS)- hope	Positive and Negative Affect Schedule (PAAS)-well-being		
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Siller et al. [2014]	USA-HIC	Cross-sectional	Caregivers of children with Autism, N=70	Mullen Scales of Early Learning (MSEL) -child's cognition	Parents' Sense of Competence Scale (PSCS)- parental competence			
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Rickards et al. [2009]	Australia-HIC	Random controlled trial	Parents of children with autism and developmental delay, N=59	Vineland Adaptive Behavior Scale (VABS)- child behaviour	Mental Development Index (MDI)- cognition	Bayley Behaviour Rating Scale-behaviour		

Questionnaire on Resources and Stress-Short Form (QRS-SF)	Lloyd & Hastings [2009]	UK- HIC	Longitudinal	Caregivers of a child with intellectual disability, N=91	Vineland Adaptive Behavior Scale (VABS)- child behaviour	Strengths and difficulties questionnaire (SDQ)- child's behaviour	Parental Locus of Control Scale (PLOC) - locus of control	Hospital Anxiety and Depression Scale (HADS) - anxiety & depression	Positive Contributions Scale (PCS) - maternal perceptions
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Gallagher et al. [2010]	UK- HIC	Cross-sectional	Caregivers of children with developmental disabilities, N=109	Strengths and difficulties questionnaire (SDQ)- child's behaviour	Pittsburgh Sleep Quality Index- Sleep quality	Family Support Scale (FSS)- family support		
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Estetes et al. [2009]	USA-HIC	Longitudinal	Caregivers of children with Autism & developmental delay, N=73	Brief Symptom Inventory (BSI)- distress	Aberrant Behavior Checklist (ABC)- child's behaviour	Vineland Adaptive Behavior Scale (VABS)- child behaviour		
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Osborne et al. [2008]	UK- HIC	Quasi-experimental	Caregivers of children with Autism, N=65	Gilliam Autism Rating Scale (GARS)- behaviour	Psycho-educational Profile—Revised (PEP-R)- capabilities	British Abilities Scale (BAS II) - cognitive abilities	Vineland Adaptive Behavior Scale (VABS)- child behaviour	
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Estes et al. [2013]	USA-HIC	Random controlled trial	Mothers of toddlers with autism spectrum disorders, N=96	Brief Symptom Inventory (BSI)- distress	Aberrant Behavior Checklist (ABC)- child's behaviour	Vineland Adaptive Behavior Scale (VABS)- child behaviour		
Questionnaire on Resources and Stress-Short Form (QRS-SF)	Quinn et al. [2007]	Ireland-HIC	Quasi-experimental	Parents of children with developmental disabilities and behavioural problems, N=41	Strengths and difficulties questionnaire (SDQ)- child's behaviour	Aberrant Behavior Checklist (ABC)- child's behaviour	The general health questionnaire-12 (GHQ-12- psychological distress	Kansas parental satisfaction scale (KPSS)- parenting 2. Family Inventory of life events & changes (FILEC) - family stress	Family assessment devise (FAD)- family functioning ; Perceived Social support scale (PSSS)
Revised Burden Measure (RBM)	Silva et al. [2015]	Portugal-HIC	Cross-sectional	Caregivers of children with Asthma, N=182	WHOQOL-BREF-QoL	Brief-COPE- coping strategies			

Revised Burden Measure (RBM)	Crespo et al. [2016]	Portugal-HIC	Cross-sectional	Parents & other relatives of children with cancer N= 204	Measure of Processes of Care-20- satisfaction with care	EUROHIS-QOL-8 - HRQoL	Satisfaction with Life Scale		
Revised Burden Measure (RBM	Carona et al. [2014]	USA-HIC	Cross-sectional	Parents of children with; 1. Epilepsy n=65 2. Cerebral palsy N=91	COPE Inventory	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)	KID-SCREEN-10 Index		
Revised Burden Measure (RBM)	Carona et al. [2013]	Portugal-HIC	Cross-sectional	1.Parents with children with cerebral palsy N=93 dyads 2. Parents with children with no medical diagnosis N=117 dyads Total N=420	Satisfaction with Social Support Scale	Satisfaction with social support scale for children and adolescents	Mental health inventory- short form (MHI-5)	Strengths and difficulties questionnaire (SDQ)	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)
Revised Burden Measure (RBM)	Carona et al. [2013]	Portugal	Cross-sectional	1.Parents with children with cerebral palsy N=105 2. Parents with children with no medical diagnosis N=117	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)				
Revised Burden Measure (RBM)	Crespo et al. [2011]	Portugal-HIC	Cross-sectional	Parents with children with asthma N=97	Family Environment Scale	DISABKIDS Chronic Generic Module	KIDSCREEN-10 Index	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)	
Revised Burden Measure (RBM)	Silva et al. [2015]	Portugal-HIC	Cross-sectional	Parents with children with asthma N=279	KIDSCREEN-10 Index	DISABKIDS-37 Chronic Generic Module	Strengths and difficulties questionnaire (SDQ)	Family Relationships Index	
Stress Level of Mothers with Children with CP	Jeong et al. [2013]	Korea- HIC	Cross-sectional	Mothers of children with CP, N=181	Carolina Parents Support Scale (CPSS) -social support				

Measurement Tool (SMCP)								
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Hsieh et al. [2013]	Taiwan-HIC	Cross-sectional	Caregivers of pre-school children with developmental delays N=70	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)	The PedsQL- Health Satisfaction Questionnaire	Impact on Family Scale Questionnaire	Pediatric Outcomes Data Collection Instrument (PODCI)
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Hsieh et al. [2016]	Taiwan-HIC	Quasi-experimental	Parents with children with developmental delays N=32	The PedsQL- Healthcare Satisfaction Module	Hospital Anxiety and Depression Scale (HADS)	World Health Organization Quality of Life Assessment Questionnaire (WHOQOL-BREF)	
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Kaugars et al. [2018]	Wisconsin, USA	Correlational	Parents with children with congenital heart disease N=54	Pediatric Inventory for Parents (PIP)	The Pediatric Quality of Life Inventory (PedsQL) Family Impact Module		
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Chen et al. [2011]	China- HIC	Cross-sectional	Parents of children with asthma n=139 & heart disease, n=264				

The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Scarpelli et al. [2008]	Brazil-UMIC	Cross-sectional	Caregivers of children with cancer N=95	Ad-hoc- clinical and demographics questionnaire				
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Appleton et al. [2012]	UK- HIC	Randomised Control Trial	Caregivers of children with neurodevelopmental problems N=275	Children's Sleep Habits Questionnaire	Epworth Sleepiness Scale	Aberrant Behaviour Checklist	Composite Sleep Disturbance Index (CSDI)	Salivary Melatonin assay
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	Hsieh et al. [2013]	Taiwan-HIC	Cross-sectional	1.Parents of preschool going children with unclassified developmental delays, n=60 2. Parents of preschool TDC, n=56	The Paediatric outcomes data collection instrument (PODCI)child's health status	WHOQOLBREF- parents' QOL	The Hospital anxiety depression scale (HADS)- parental psychological distress	The PedsQL Health satisfaction- parental satisfaction with the child's health care	
The Pediatric Quality of Life Inventory- Generic Core Scales (PedsQL)- Family Impact Module	M. van der Holst et al. [2016]	Netherlands-HIC	Cross-sectional	Parents of children with neonatal brachial plexus palsy, N=59	TNO-AZL Preschool children quality of life (TAP QOL) - quality of life	21 Upper extremity functioning questions- upper extremity function			
Zarit Burden Inventory (ZBI)	Wang et al. [2017]	China- HIC	Quasi-experimental	Parents of children with lymphoblastic leukaemia, N=130	Zung's Self-Rating Anxiety Scale (SAS)- Anxiety	Zung's Self-Rating Depression Scale (SDS)- depression	Perceived Social Support Scale (PSSS)- social support	Medical Outcomes Study 36-item Short Form (SF-36)- HRQoL	

Zarit Burden Inventory (ZBI)	Suzuki et al. [2014]	Japan-HIC	Random controlled trial	Mothers of children with high-functioning pervasive developmental disorders, N=72	GHQ-28- psychiatric morbidity	Medical Outcomes Study 36-item Short Form (SF-36)- HRQoL	Japanese version of the ABC-behaviour	
Zarit Burden Inventory (ZBI)	Gallagher et al. [2008]	Britain - HIC	Cross-sectional	Parents of Children with Intellectual Disabilities, N=61	Hospital Anxiety and Depression Scale (HADS) - anxiety & depression	Support Functions Scale (SFS) - social support	Strengths and Difficulties Questionnaire (SDQ)- child behavioural problems	Pittsburgh Sleep Quality Index- Sleep quality
Zarit Burden Inventory (ZBI)	Pedrón-Giner et al [2014]	Spain-HIC	Cross-sectional	Caregivers of children with neurological disease and home enteral nutrition, N=58	Gross Motor Function Classification System (GMFCS)- functioning	Symptom Checklist 90 Revised- distress		
Zarit Burden Inventory (ZBI)	Ikeda et al. [2012]	Japan-HIC	Cross-sectional	Caregivers of children with physical disabilities, N=100	Pittsburgh Sleep Quality Index- Sleep quality	Child sleep problems		
Zarit Burden Inventory (ZBI)	Landfeldt et al. [2016]	Germany, Italy &USA-HICs	Cross-sectional	Caregivers to patients with Duchenne Muscular Dystrophy, N=770	EuroQol EQ-5D-3L	SF-12 Health Survey (SF12)		

11.2 Appendix 2: ZCCS item bank gleaned from the systematic review

Physical Burden		
Outcome measure	**Subtheme**	**Item**
CSI & MCSI	Summative	In general, caregiving is a physical strain (For example: lifting in or out of a chair; effort or concentration is required)
Care-ILI-QOL	Sleep	It is difficult to get sleep at night
CSI & MCSI	Sleep	disruption of sleep
CSI & MCSI	Sleep	My sleep is disturbed (For example: the person I care for is in and out of bed or wanders around at night)
BSFC	physical	Sleep deprivation
CGSQ	Tiredness	Feeling tired or strained
KVCAS	Tiredness	Do you feel tired as a result of caring for child?
Caregiver difficulties Scale (CDS)	Tiredness	Does caring for the child make you feel tired and exhausted?
CGSQ	Pain	I feel body aches when lifting and providing for care
Economic burden		
Outcome measure	**Subtheme**	**Item**
Caregiver difficulties Scale (CDS)	Expenses	Is there an increase in your family expenses due to the child's condition?
caregiver burden scale V2	Expenses	Spending a large amount of money
Caregiver difficulties Scale (CDS)	Adequacy	Is your income adequate to provide the necessities for the child?
Zarit Burden Interview	Adequacy	Do you feel that you don't have enough money to take care of your child in addition to the rest of your expenses?

caregiver burden scale V2	Adequacy	Our finances are not able to take care of other family members
CGSQ	Adequacy	Any family member having to do without things
Caregiver difficulties Scale (CDS)	economic	Do you worry that you are unable to provide special facilities and services needed by your child?
CSI & MCSI	Work adj.	There have been work adjustments (For example, I have to take time off for caregiving duties)
CGSQ	Work adj.	Missing work or neglecting other duties
CSI & MCSI	Overall	Caregiving is a financial strain
APSOM	Overall	My child's condition places a financial strain on my family
PSI-SF		I am unhappy with the last purchase of clothing I made for myself
Impact on Family		
Outcome measure	**Subtheme**	**Item**
CGSQ	Attention	Less attention paid to other family members
Caregiver difficulties Scale (CDS)	Attention	Does the child's condition prevent you from attending to the needs of other family members?
caregiver burden scale V1	Attention	Have your caregiving responsibilities: Given you little time for friends and relatives?
caregiver burden scale V2	Attention	Difficulty in keeping contact with relatives and friends
caregiver burden scale V2	Relationships	Worse relationship with family members
CGSQ	Relationships	Disruption or upset of relationships within the family
Zarit Burden Interview	Relationships	Do you feel that your relative currently affects our relationships with other family members or friends in a negative way?
caregiver burden scale V1	Relationships	Have your caregiving responsibilities: Caused conflicts with your relatives?

CSI & MCSI	Adjustments	There have been family adjustments (For example: helping has disrupted my routine; there is no privacy)
CGSQ	Adjustments	Disruption of family routines
CGSQ	Adjustments	Disruption of family' social activities
caregiver burden scale V2	Adjustments	The whole family having to readapt themselves
APSOM	Psychological	I feel guilty for spending more time with my child with a medical condition than my other family members
APSOM	Psychological	Family members resent the time I spend with my child
Zarit Burden Interview	Psychological	Do you feel stressed between caring for your relative and trying to meet other responsibilities for your family or work?
caregiver burden scale V2	Psychological	Blamed by other family members for not taking good care of the child
APSOM	Positive influence	My child's condition has brought my family closer together
CGSQ	Future	Feeling worried about family's future
CGSQ	Overall	Toll taken on family
APSOM	Overall	My family has been significantly impacted by my child's condition
Support systems		
Outcome measure	**Subtheme**	**Item**
Care-ILI-QOL	Family	I do get practical support from my family
Care-ILI-QOL	Family	I do get emotional support from my family?
CDS	Family	Are you able to discuss your child's problems with other family members?
Care-ILI-QOL	Friends	I do get emotional support from my friends?
CDS	Community	Do your relatives/neighbours help you with caring for the child?

CDS	Spouse	Does your spouse help you with the care of this child?
CDS	Spouse	Does your spouse support you in other family responsibilities?
PECI	Overall	I can get help and support when I need it
APSOM	Overall	I feel like I have adequate support caring for my child
Concerns for child		
Outcome measure	**Subtheme**	**Item**
Caregiver difficulties Scale (CDS)	child	Does your child fall ill from time to time?
Caregiver difficulties Scale (CDS)	child	Are you satisfied about the improvement in your child's condition after receiving treatment/therapy?
Caregiver difficulties Scale (CDS)	child	Do you fear what your child's future might be?
Caregiver difficulties Scale (CDS)	child	Do you worry about your child's present state?
Caregiver difficulties Scale (CDS)	child	Do you worry that your child cannot function like other children (e.g. going to school, playing)?
Caregiver difficulties Scale (CDS)	child	Do you feel sad that your child cannot do anything by him/herself?
Caregiver difficulties Scale (CDS)	child	Do you worry that your child gets insulted and/or ridiculed by others?
Caregiver difficulties Scale (CDS)	child	Do you fear that your child will have accidents because of his/her disability?
CSI & MCSI	child	Some behaviour is upsetting (For example: incontinence; cries a lot)
PSI-SF (parenting stress index)	child	There are some things my child does that really bother me a lot.
PSI-SF (parenting stress index)	child	My child generally wakes up in a bad mood
PSI-SF (parenting stress index)	child	I feel that my child is very moody and easily upset
PSI-SF (parenting stress index)	child	My child does a few things that bother me a great deal
PSI-SF (parenting stress index)	child	My child reacts very strongly when something happens that my child doesn't like

PSI-SF (parenting stress index)	child	My child gets upset easily over the smallest thing
PSI-SF (parenting stress index)	child	My child's sleeping and eating schedule were much harder to establish than I expected.
PSI-SF (parenting stress index)	child	I have found that getting my child to do something is hard
PSI-SF (parenting stress index)	child	Think carefully and count the number of things which your child does that bothers you.
PSI-SF (parenting stress index)	child	My child turned out to be more of a problem than I expected.
PSI-SF (parenting stress index)	child	My child makes more demands on me than most children.
PSI-SF (parenting stress index)	child	My child seems to cry more often than most children
PSI-SF (parenting stress index)	child	When playing, my child doesn't often giggle or laugh
PSI-SF (parenting stress index)	child	It takes a long time, and it is really hard for my child to get used to new things
caregiver burden scale V1	child	Increased attempts by your child to manipulate you?
Zarit Burden Interview	child	Are you afraid what the future holds for your child?
Zarit Burden Interview	child	Do you feel embarrassed over your child's behaviour?
APSOM	child	I feel a sense of loss when I think about my child's future
APSOM	child	I worry about my child's future more than other parents because of his and/or her medical condition
caregiver burden scale V2	child	Embarrassed about child's behaviour
Care-ILI-QOL	child	I am worried about bringing my sick child out to meet other people?
CGSQ	child	difficulty in relating to child
CGSQ	child	Feeling worried about the child's future
autism PSI	child	concern for the future of your child being accepted by others
autism PSI	child	concern for the future of your child living independently
PECI	child	I worry about something bad happening to my child when s/he is out of my care.
PECI	child	When my child is actively playing, I find myself worried that s/he will get hurt.
PECI	child	I wake up during the night and check on my child.
PECI	child	When I'm not with my child, I find myself thinking about whether or not s/he is ok
PECI	child	I worry about my child's future.
PECI	child	I worry about whether my child will be able to live independently as an adult
PCDI	child	Others tease or call my child names.
PCDI	child	My child seems lonely.

Impact on self		
Outcome measure	**Subtheme**	**Item**
Caregiver difficulties Scale (CDS)	Time	Do you have enough time to look after your own health?
Caregiver difficulties Scale (CDS)	Time	Do you have enough time for your basic daily needs such as having meals, sleeping, bathing etc.?
Caregiver difficulties Scale (CDS)	Time	Do you feel that you will never have enough time to get everything done?
CSI & MCSI	time	There have been other demands on my time (For example: other family members need me)
caregiver burden scale V1	Time	Have your caregiving responsibilities: Decreased time you have to yourself?
Zarit Burden Interview	Time	Do you feel that because of the time you spend with your child that you don't have enough time for yourself?
CGSQ	Time	Interruption of personal time
BSFC	Time	Not enough time for my own interests
KRCAS	Time	I can fit in most of the things I need to do in spite of the time it takes to care for child
BICSF	Time	decreased time for yourself
Caregiver difficulties Scale (CDS)	Health	Do you think that your health has been affected because of your child's condition?
Zarit Burden Interview	Health	Do you feel your health has suffered because of your involvement with your child?
KRCAS	Health	I feel that my health has suffered because of the care I give to the child
BICSF	Health	my body aches when providing for care
CSI & MCSI	Restrictive	My sleep is disturbed (For example: the person I care for is in and out of bed or wanders around at night)
CSI & MCSI	Restrictive	Caregiving is inconvenient (For example: helping takes so much time)
CSI & MCSI	Restrictive	There have been changes in personal plans (For example: I had to turn down a job)

APSOM	Restrictive	My child's condition limits my personal activities
PSI-SF (parenting stress index)	Restrictive	I feel trapped by my responsibilities as a parent/guardian
BICSF	Restrictive	I cannot freely leave the house because of caregiving
PSI-SF (parenting stress index)	Restrictive	I find myself giving up more of my life to meet my child's needs than I ever expected
PECI	Self-efficacy	I trust myself to manage the future, whatever happens
PSI-SF (parenting stress index)	Self-efficacy	I often have the feeling that I cannot handle things very well
Zarit Burden Interview	Self-efficacy	Do you feel you have lost control of your life since your child's illness?
caregiver burden scale V2	Future	Difficulty in planning for the future
PECI	Future	I feel ready to face challenges related to my child's well-being in the future
PSI-SF (parenting stress index)	stress	I am unhappy with the last purchase of clothing I made for myself
PSI-SF (parenting stress index)	Stress	I am not as interested in people as I used to be
PSI-SF (parenting stress index)	stress	I don't enjoy things as I used to.
APSOM	Marriage	Parenting a child with a medical condition places strain on marriages and common-law relationships
APSOM	Other	Having a child with a medical condition makes the decision to have more children difficult
Impact on Social life		
Outcome measure	**Subtheme**	**Item**
Caregiver difficulties Scale (CDS)	confining	Do you have to restrict your social visits and relationships due to the child's illness?
CSI & MCSI	confining	Caregiving is confining (For example: helping restricts free time, or I cannot go visiting)
caregiver burden scale V2	confining	Everyday activities dependent on recipient's need

caregiver burden scale V2	confining	Unable to go on a trip
CQOLC	confining	unable to attend church/religious activities
PSI-SF (parenting stress index)	social	Since having my child, I have been unable to try new and different things
PSI-SF (parenting stress index)	social	Since having my child, I feel that I am almost never able to do things that I like to do
PSI-SF (parenting stress index)	social	I feel alone and without friends
Zarit Burden Interview	social	Do you feel uncomfortable about having friends over because of your child?
DAS21	social	I find it difficult to relax
PSI-SF (parenting stress index)	Marriage	Having a child has caused more problems than I expected in my relationship with my spouse
caregiver burden scale V1	recreation	Kept you from recreational activities?
Caregiver difficulties Scale (CDS)	recreation	Does the child's condition prevent you from being relaxed?
caregiver burden scale V1	recreation	Left you with almost no time to relax?
caregiver burden scale V1	Summative	Caused your social life to suffer?
Zarit Burden Interview	Summative	Do you feel that your social life has suffered because you are caring for your child?
Overall burden perception		
Outcome measure	**Subtheme**	**Item**
Zarit Burden Interview	overall	Overall, how burdened do you feel in caring for your child?
BICSF	overall	I am completely distressed by caregiving
CSI & MCSI	overall	I feel completely overwhelmed (For example: I worry about the person I care for; I have concerns about how I will manage)

11.3 Appendix 3: Caregiver Interview Guide

Caregiver Unique Identification Number_____

Venue: _____

Date of interview: _____

Interview start time: _____ Interview end time: _____

a. Warm-up questions:

- Can you please tell me more about yourself?
- Background prior to assuming the caregiving role?
- What is a typical day like for you?

Note: start with warm-up questions to create trust and rapport with the prospective participant. Adopt questions depending on the participants' responses.

b. Caregiving challenges

Theme	Questions
Concerns for the child	• What your concerns/ worries towards the child's condition, e.g. concerns about the future. NB- allow the caregiver to express themselves fully. • Thereafter, ask probing questions to elicit more information, an example of probes include: worries on physical development, future of the child, etc.
Physical challenges	• Do you face challenges in lifting and transfers? • Do you feel some bodily pain in the process of care? • Do you feel that your health has been affected by caregiving? •
Economic challenges	• Have there been changes in your family expenses because of the needs of the child? • Follow up question - If there has been an increase, can you please further elaborate? • Have there been alterations in your income-generation activities due to the caregiving role? •
Community participation	• Have you faced any challenges within the community, e.g. stigma? • Have your social relations changed because of the caregiving role? •
Family	• Have there been changes/challenges with your spouse because of the disabled child? • Have there been any changes in your immediate family dynamics? • Has the condition of your child affected your relationship with the extended family, i.e. in-laws in particular?

c. Closing questions

Before concluding the interview, ask the following questions to elicit additional views which may not have been succinctly covered by the interview guide:

- "Is there anything you would like to add?"
- "What else should we talk about regarding caregiving challenges?"

Notes:

I. Start with more general and progress to more specific questions
II. Give the respondent time to reflect and then respond to the questions
III. Were appropriate, seek clarity by asking a follow-up and or probing questions, e.g. Can you give me an example of what you mean?
IV. Allow the respondent to express themselves emotionally
V. Make use of voice variation, gestures and body language to demonstrate an interest in the caregivers' narrative
VI. Acknowledge contributions from the participants, e.g. What you are sharing (or have said) is essential. Can you say more?
VII. Conclude interview by thanking the participant!

11.4 Appendix 4: ZCCS Version 2.0 – 80 items for experts' evaluation

#	Domain	Sub-Theme	Question	Not relevant	Somewhat relevant	Quite relevant	Highly relevant	Additional comments
1.	Self	Health	Considering your caregiving responsibilities, do you have enough time for your basic needs such as having meals, sleeping, bathing, etc.?	1	2	3	4	
2.	Self	Health	In general, I feel that my health has suffered because of the care I provide to the child.	1	2	3	4	
3.	Self	Health	When I compare my general level of health over the past 12 months, I feel that my state of health today has worsened?	1	2	3	4	
4.	Self	Time	I can fit in most of the things I need to do in spite of the time it takes to care for the child?	1	2	3	4	
5.	Self	Restrictive	I feel trapped by my responsibilities as a parent/guardian.	1	2	3	4	
6.	Self	Restrictive	I find myself giving up more of my life to meet my child's needs than I ever expected	1	2	3	4	
7.	Self	Self-efficacy	Do you feel you have lost control of your life because of caregiving?	1	2	3	4	
8.	Self	Self-efficacy	I feel ready to face challenges related to my child's well-being in the future.	1	2	3	4	
9.	Self	Self-efficacy	In general, I often have the feeling that I cannot handle things very well.	1	2	3	4	
10	Self	Self-efficacy	Due to the responsibilities of caregiving, it is now difficult to plan for the future?	1	2	3	4	
11	Self	Other	Having a child with a disability makes the decision to have more children difficult.	1	2	3	4	
12	Physical	Sleep	My sleep is disturbed (For example: the child I care for cries a lot and wakes me up at night)	1	2	3	4	

13	Physical	Exhaustion	I feel tired and exhausted as a result of caring for the child.	1	2	3	4	
14	Physical	Pain	I feel body aches or discomfort when providing for care.	1	2	3	4	
15	Physical	Overall	In general, caregiving is a physical strain, i.e. it requires a lot of physical effort in performing the caregiving roles	1	2	3	4	
16	Economic	Work	There have been changes in work plans due to caregiving (For example: I had to turn down a job)	1	2	3	4	
17	Economic	Work	There have been work adjustments due to caregiving (For example: I have to take time off for caregiving duties)	1	2	3	4	
18	Economic	Expenses	Has there been an increase in your family expenses due to the child's condition?	1	2	3	4	
19	Economic	Worry	Do you worry that you are unable to provide special facilities and services needed by your child?	1	2	3	4	
20	Economic	Adequacy	Is your income adequate to provide the necessities for the child?	1	2	3	4	
21	Economic	Adequacy	Is your income adequate able to take care of other family members?	1	2	3	4	
22	Economic	Overall	In general, I feel that caregiving is a financial strain	1	2	3	4	
23	Family	Attention	Does providing for care prevent you from attending to the needs of other family members?	1	2	3	4	
24	Family	Relationships	There has been a disruption or upset of relationships within the family?	1	2	3	4	
25	Family	Relationships	Parenting a child with a disability has caused more problems than I expected in my relationship with my spouse (significant other)	1	2	3	4	
26	Family	Adjustments	There have been adjustments/disruptions in family routines and social activities?	1	2	3	4	
27	Family	Psychological	Family members resent the time I spend with my child	1	2	3	4	

28	Family	QOL	I feel that caregiving has decreased our family standard of living, for example, some family members had to do without basic necessities.	1	2	3	4	
29	Family	Overall	Overall, I feel that my family has been negatively affected by my child's condition.	1	2	3	4	
30	Child	Future	I worry about my child's future more than other parents because of his/her disability	1	2	3	4	
31	Child	Future	I worry about whether my child will be able to live independently as an adult	1	2	3	4	
32	Child	Behaviour	I feel embarrassed about my child's behaviour?	1	2	3	4	
33	Child	Behaviour	My child seems to cry more often than most children	1	2	3	4	
34	Child	Behaviour	Do you feel that your child asks for more help than he/she needs?	1	2	3	4	
35	Child	Behaviour	My child gets upset easily over the smallest thing.	1	2	3	4	
36	Child	Stigma	Do you worry that your child gets insulted and/or ridiculed by others?	1	2	3	4	
37	Child	Stigma	I am worried about bringing my child out to meet other people?	1	2	3	4	
38	Child	Stigma	Other people don't know how to treat my child.	1	2	3	4	
39	Child	Stigma	Do you have to face embarrassing situations when you are travelling with the child?	1	2	3	4	
40	Child	functioning	Do you feel sad that your child cannot do anything by him/herself like other children (e.g. going to school, playing)?	1	2	3	4	
41	Child	functioning	My child is not able to do as much as I expected.	1	2	3	4	
42	Child	Parental worry	I worry about something bad happening to my child when s/he is out of my care.	1	2	3	4	

43	Child	Parental worry	I wake up during the night and check on my child.	1	2	3	4	
44	Child	Other	My child's sleeping and eating schedule were much harder to establish than I expected.	1	2	3	4	
45	Child	Health	My child falls ill from time to time?	1	2	3	4	
46	Child	Other	Are you satisfied with the improvement in your child's condition after receiving treatment/therapy?	1	2	3	4	
47	Social support	Family	I do get practical support from my family.	1	2	3	4	
48	Social support	Family	I do get emotional support from my family.	1	2	3	4	
49	Social support	Family	Are you able to discuss your child's problems with other family members?	1	2	3	4	
50	Social support	Friends	Do your friends help you with caring for the child?	1	2	3	4	
51	Social support	Community	Do your neighbours help you with caring for the child?	1	2	3	4	
52	Social support	Spouse	Does your spouse help you with the care of this child?	1	2	3	4	
53	Social support	Spouse	Does your spouse support you in other family responsibilities?	1	2	3	4	
54	Social support	Overall	Overall, I feel like I have adequate help and support caring for my child	1	2	3	4	

55	Psychological	Emotions	I expected to have closer and warmer feelings for my child than I do and this bothers me	1	2	3	4	
56	Psychological	Anger	I feel anger when I think about the potential cause(s) of my child's condition	1	2	3	4	
57	Psychological	Anger	I experience angry feelings when I think about my child's illness	1	2	3	4	
58	Psychological	Self-efficacy	Do you feel uncertain about what to do about your child?	1	2	3	4	
59	Psychological	Self-efficacy	Do you feel you should be doing more for your child?	1	2	3	4	
60	Psychological	Self- efficacy	When I do things for my child, I get the feeling that my efforts are not appreciated very much	1	2	3	4	
61	Psychological	Self-efficacy	I feel confident in about my ability to handle problems associated with caring for a child with a disability	1	2	3	4	
62	Psychological	Guilt	I worry that something I did or my partner did during pregnancy caused my child's condition	1	2	3	4	
63	Psychological	Guilt	I feel guilty about the potential cause(s) of my child's condition	1	2	3	4	
64	Psychological	Aetiology	I am focused on finding a specific reason for why this happened to my child	1	2	3	4	
65	Psychological	Aetiology	My child's condition could have been prevented	1	2	3	4	
66	Psychological	Aetiology	My child's condition was caused by witchcraft, or a is a sign of bad luck/omen	1	2	3	4	
67	Psychological	Aetiology	I have come to terms with my child's condition	1	2	3	4	
68	Psychological	Sad	Seeing healthy children doing everyday activities makes me feel sad.	1	2	3	4	
69	Psychological	Locus	I feel that I have lost control of my life because of caring for child?	1	2	3	4	
70	Psychological	Stress	I find myself to be easily agitated	1	2	3	4	
71	Psychological	Hopelessness	Having caregiving responsibilities has created a feeling of hopelessness?	1	2	3	4	

72	Social life	Confining	Because of caregiving, it is difficulty in keeping contact with relatives and friends?	1	2	3	4	
73	Social life	Isolation	I feel isolated and alone as a result of caring for child					
74	Social life	Time	Do you feel that because of the time you spend with your child that you don't have enough time for your own interests/hobbies?					
75	Social life	Hobbies	Since having my child, I have been unable to try new and different things	1	2	3	4	
76	Social life	Summative	In general, do you feel that your social life has suffered because of providing care to your child?	1	2	3	4	
77	Overall	Overall	I feel completely overwhelmed by the caregiving role.	1	2	3	4	
78	Overall	Overall	I am unable to care for child much longer	1	2	3	4	
79	Overall	Overall	I have had more negative than positive experiences parenting a child with cerebral palsy	1	2	3	4	
80	Overall	Overall	I wish if I could leave the care of the child to someone else?	1	2	3	4	

11.5 Appendix 5: UCT HREC approval letter

UNIVERSITY OF CAPE TOWN
Faculty of Health Sciences
Human Research Ethics Committee

Room E52-24 Old Main Building
Groote Schuur Hospital
Observatory 7925
Telephone [021] 406 6338
Email: lamees.emjedi@uct.ac.za
Website: www.health.uct.ac.za/fhs/research/humanethics/forms

05 May 2016

HREC REF: 122/2016

Prof J Jelsma
Health & rehabilitation Sciences
Physiotherapy
F45, OMB

Dear Prof Jelsma

PROJECT TITLE: DETERMINATION OF THE IMPACT OF REHABILITATION VILLAGES AS A SERVICE DELIVERY MODEL: A STUDY OF VIEWS AND EXPERIENCES OF ZIMBABWEAN CAREGIVERS OF CHILDREN WITH CEREBRAL PALSY (PhD candidate - Jermaine Dambi)

Thank you for submitting your response to the Faculty of Health Sciences Human Research Ethics Committee dated 19 April 2016.

It is a pleasure to inform you that the HREC has **formally approved** the above-mentioned study.

Approval is granted for one year until the 30th May 2017.

The HREC acknowledge that the following PhD candidate - Jermaine Dambi will also be involved in this study.

Please submit a progress form, using the standardised Annual Report Form if the study continues beyond the approval period. Please submit a Standard Closure form if the study is completed within the approval period.
(Forms can be found on our website: www.health.uct.ac.za/fhs/research/humanethics/forms)

Please note that the ongoing ethical conduct of the study remains the responsibility of the principal investigator.

Please note that for all studies approved by the HREC, the principal investigator **must** obtain appropriate institutional approval before the research may occur.

Please quote the HREC REF in all your correspondence.

Yours sincerely

PROFESSOR M BLOCKMAN
CHAIRPERSON, FHS HUMAN RESEARCH ETHICS COMMITTEE
Federal Wide Assurance Number: FWA00001637.

11.6 Appendix 6: MRCZ approval letter

Telephone: 791792/791193
Telefax: (263) - 4 - 790715
E-mail: mrcz@mrcz.org.zw
Website: http://www.mrcz.org.zw

Medical Research Council of Zimbabwe
Josiah Tongogara / Mazoe Street
P. O. Box CY 573
Causeway
Harare

APPROVAL

REF: MRCZ/A/2072 **04 July 2016**

Dambi Jermaine Matewu
University of Zimbabwe
College of Health Sciences
Department of Rehabilitation
P.O Box A 178
Avondale
Harare

RE:-Determination of the impact of rehabilitation villages as a service delivery model:- A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Thank you for the application for reviewof Research Activity that you submitted to the Medical Research Council of Zimbabwe (MRCZ). Please be advised that the Medical Research Council of Zimbabwe has **reviewed** and **approved** your application to conduct the above titled study.

Thisapproval is based on the review and approval of the following documents that were submitted to MRCZ for review:-

a) Full Proposal
b) Informed Consent Forms (English and Shona)
c) Data Collection Tools (English and Shona)

- **APPROVAL NUMBER : MRCZ/A/2072**

This number should be used on all correspondence, consent forms and documents as appropriate.

- **TYPE OF MEETING : Expedited**
- **EFFECTIVE APPROVAL DATE : 04 July 2016**
- **EXPIRATION DATE : 03 July 2017**

After this date, this project may only continue upon renewal. For purposes of renewal, a progress report on a standard form obtainable from the MRCZ Offices should be submitted three months before the expiration date for continuing review.

- **SERIOUS ADVERSE EVENT REPORTING**: All serious problems having to do with subject safety must be reported to the Institutional Ethical Review Committee (IERC) as well as the MRCZ within 3 working days using standard forms obtainable from the MRCZ Offices or website.
- **MODIFICATIONS**: Prior MRCZ and IERC approval using standard forms obtainable from the MRCZ Offices is required before implementing any changes in the Protocol (including changes in the consent documents).
- **TERMINATION OF STUDY**: On termination of a study, a report has to be submitted to the MRCZ using standard forms obtainable from the MRCZ Offices or website.
- **QUESTIONS**: Please contact the MRCZ on Telephone No. (04) 791792, 791193 or by e-mail on mrcz@mrcz.org.zw

Other

- Please be reminded to send in copies of your research results for our records as well as for Health Research Database.
- You're also encouraged to submit electronic copies of your publications in peer-reviewed journals that may emanate from this study.

Yours Faithfully

.....

MRCZ SECRETARIAT
FOR CHAIRPERSON
MEDICAL RESEARCH COUNCIL OF ZIMBABWE

MEDICAL RESEARCH COUNCIL OF ZIMBABWE
2016 -07- 04
APPROVED
P.O. BOX CY 573 CAUSEWAY, HARARE

PROMOTING THE ETHICAL CONDUCT OF HEALTH RESEARCH

11.7 Appendix 7: Consent forms

11.7.1 Panel of experts' consent form

IRB No. _____

School of Health and Rehabilitation Sciences

Faculty of Health Sciences

Divisions of Communications Sciences and Disorders, Disability Studies, Nursing and Midwifery, Occupational Therapy, Physiotherapy

F45 Old Main Building, Groote Schuur Hospital,

Observatory 7925

Tel: +27 (0) 21 406 6401 Fax: +27 (0) 21 406 6323

Internet: www.uct.ac.za

Study title: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Principal Investigator: Dambi Jermaine, [Msc. PT, Bsc. (Hons) PT]

Co-Investigator(s): Professor J Jelsma, Mrs T Mlambo

Phone number(s): 0773 444 911

What you should know about this research study:

- We give you this consent so that you may read about the purpose, risks, and benefits of this research study.
- Routine care is based upon the best-known treatment and is provided with the main goal of helping the individual patient. The main goal of research studies is to gain knowledge that may help future patients.
- We cannot promise that this research will benefit you. However, we aim to develop an outcome measure which may improve the efficacy of management of life-long disabilities
- You have the right to refuse or agree to participate in taking part now and changing your mind later.
- Whatever you decide, it will not affect you in any way.
- Please review this consent form carefully. Ask any questions before you make a decision.
- You are free not to answer any questions
- Your choice for participation is voluntary.

PURPOSE

Provision of rehabilitation services in Zimbabwe is mainly through the hospital-based approach, and some hospitals additionally offer rehabilitation village–based treatment services for children with cerebral palsy (disabilities). However, nothing is known about the best method of delivering rehabilitation services. This study hopes to come up with recommendations of the most effective model for the treatment of children with cerebral palsy (CP) and providing psychosocial care and support to caregivers as long-term caregiving has been shown to have a negative effect on the health and well-being of the caregiver. Thus, the findings of this research will be used to come with recommendations for a model that provides holistic treatment. However, the evaluation of the efficacy of the models is very much dependent using valid and reliable outcome measures. Unfortunately, most tools which measure patient-reported health outcomes had been developed in high-income countries and their applicability in low resources settings may be questionable due to differences in culture and socio-economic backgrounds. We will, therefore, develop a context-specific and culturally relevant caregiver burden outcome measure. We will also adapt, translate and validate the multidimensional social support scale (MSPPS) into Shona since there are no tools for measuring social support validated for use in the local context. It is important to evaluate social support level as it has been shown that social support is an essential coping mechanism for caregivers.

PROCEDURES AND DURATION

If you decide to participate, you will be requested to assess the face validity of the ZCCS questionnaire by giving your views on the relevancy of the preliminary pool of items. We anticipate this phase of the study to take up to a week depending on the response from other panel members between two to four weeks. In addition, you will not benefit directly from participation, but it's the hope of the researcher that the outcomes of this study will assist in coming up with evidence-based recommendations of a model which will ultimately assist in the planning of rehabilitation services and improving treatment efficacy.

RISKS AND DISCOMFORTS

This study will not pose any danger to you except that we may require some of your time which may disrupt your daily schedule on the days of scheduled meetings.

BENEFITS AND/OR COMPENSATION

There are no associated benefits in participating in this study. However, you will be provided with food and drinks after the meetings. Additionally, should require any further treatment and support, the researcher will be glad to assist you and or refer you to other people who can assist you.

CONFIDENTIALITY

Information gathered will be kept in private. You will be assigned a code/number for identification purposes, and no names will be used for reference in the study and publications. The audio recordings of the interviews will be burnt onto DVDs and will be kept in a safe and locked drawer at The University of Zimbabwe. The recordings will be burnt a year after. We will inform you about the outcomes of the present study through an oral presentation and leaflets with summarized information at the end of the study. In addition, only the researcher and under some circumstances, the MRCZ may need to review the audio recordings for compliance audits

 IRB No. _____

ADDITIONAL COSTS

There are no additional costs associated with your participation in this study.

VOLUNTARY PARTICIPATION

Please do take note that you are not obliged to participate in the study, i.e. participation is on a voluntary basis. You will be free to withdraw from the study at any stage without any penalty or need to explain your withdrawal. However, if you choose to withdraw, please do try by all means to notify the researcher about your decision.

SIGNATURE PAGE

Study title: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Protocol Version Number/date

OFFER TO ANSWER QUESTIONS

Before you sign this form, please ask any questions on any aspect of this study that is unclear to you. You may take as much time as necessary to think it over.

AUTHORIZATION

You are making a decision whether or not to participate in this study. Your signature indicates that you have read and understood the information provided above, have had all your questions answered, and have decided to participate.

_____ _____

Name of Research Participant (please print) Date

_____ _____

Signature of Participant Time

_____ _____

Name of Staff Obtaining Consent Signature & Date

YOU WILL BE OFFERED A COPY OF THIS CONSENT FORM TO KEEP.

If you have any questions concerning this study or consent form beyond those answered by the investigator, including questions about the research, your rights as a research participant or research-related injuries; or if you feel that you have been treated unfairly and would like to talk to someone other than a member of the research team, please feel free to contact the Medical Research Council of Zimbabwe (MRCZ) on telephone (04)791792 or (04) 791193 and cell phone lines 0784 956 128. The MRCZ Offices are located at the National Institute of Health Research premises at Corner Josiah Tongogara and Mazowe Avenue in Harare.

11.7.2 Caregivers consent form – Phase 1 (English version)

 IRB No. _____

School of Health and Rehabilitation Sciences

Faculty of Health Sciences

Divisions of Communications Sciences and Disorders, Disability Studies, Nursing and Midwifery, Occupational Therapy, Physiotherapy

F45 Old Main Building, Groote Schuur Hospital,

Observatory 7925

Tel: +27 (0) 21 406 6401 Fax: +27 (0) 21 406 6323

Internet: www.uct.ac.za

Study title: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Principal Investigator: Dambi Jermaine, [Msc. PT, Bsc. (Hons) PT]

Co-Investigator(s): Professor J Jelsma, Mrs T Mlambo

Phone number(s): 0773 444 911

What you should know about this research study:

- We give you this consent so that you may read about the purpose, risks, and benefits of this research study.
- Routine care is based upon the best-known treatment and is provided with the main goal of helping the individual patient. The main goal of research studies is to gain knowledge that may help future patients.
- We cannot promise that this research will benefit your child. Just like regular care, this research can have side effects that can be serious or minor.
- You have the right to refuse to allow your child to take part, or agree for your child to take part now and change your mind later.
- Whatever you decide, it will not affect your child's regular care.
- Please review this consent form carefully. Ask any questions before you make a decision.
- You are free not to answer any questions
- Your choice to allow your child to participate is voluntary.

PURPOSE

They are different ways of providing rehabilitation services. One method is whereby clients walk into a hospital for rehabilitation services straight from their homes. However, some hospitals offer services through rehabilitation villages. These are housing structures whereby caregivers and children with disabilities camp at the district hospitals for up to three days. Their children get rehabilitation treatment during the stay. However, nothing is known about the best method of delivering rehabilitation services. This study hopes to find the best way of providing rehabilitation services as well as ways of providing support for caregivers. This is because people who care for children with disabilities may face certain challenges in looking after these children. It is against this background that we will interview you and then develop a questionnaire to try to understand your experiences in taking of a child with cerebral palsy (CP). Thus, the findings of this research will be used to develop ways to assist caregivers in coping with the challenges faced in providing care to a child with a disability. Since you have a child with CP, we would like to invite you to take part in our study.

PROCEDURES AND DURATION

If you decide to participate, we will interview you to understand your experiences in providing for care to a child with a disability. We expect the interviews to take between 30 minutes to an hour. We will also kindly ask you to fill in the questionnaire so that you may also give us feedback as to whether the questions are appropriate or not. This phase of the study is expected to run for two to four weeks.

RISKS AND DISCOMFORTS

This study will not pose any danger to you except that we may require some of your time which may disrupt your daily schedule on the day we will ask you questions.

BENEFITS AND/OR COMPENSATION

There are no associated benefits in participating in this study. However, you (and the children) will be provided with food and drinks after the interviews and upon completion of the questionnaires. Additionally, should require any further treatment and support, the researcher will be glad to assist you and or refer you to other people who can assist you.

CONFIDENTIALITY

Information gathered about you, and your child will be kept in private. You will be assigned a code/number for identification purposes, and no names will be used for reference in the study and publications. The audio recordings of the interviews will be burnt onto DVDs and will be kept in a safe and locked drawer at The University of Zimbabwe. The recordings will be burnt a year after completion of the study. In addition, only the researcher and under some circumstances, the MRCZ may need to review patient records for compliance audits. We will inform you about the outcomes of the present study through oral presentations and leaflets with simplified information at the end of the study.

ADDITIONAL COSTS

There are no additional costs associated with your participation in this study as we will ask you questions on the days you bring your child for the usual treatments.

VOLUNTARY PARTICIPATION

Participation in this study is voluntary. If you decide not to participate in this study, your decision will not affect your future relations with the hospital and its personnel. If you decide to participate, you are free to withdraw your consent and to discontinue participation at any time without penalty. However, in the event that you choose to withdraw, please do try by all means to notify the researcher about your decision.

 IRB No. _____

SIGNATURE PAGE

Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Protocol Version Number/date

OFFER TO ANSWER QUESTIONS

Before you sign this form, please ask any questions on any aspect of this study that is unclear to you. You may take as much time as necessary to think it over.

AUTHORIZATION

You are making a decision whether or not to participate in this study. Your signature indicates that you have read and understood the information provided above, have had all your questions answered, and have decided to participate.

_____ _____

Name of Research Participant (please print/ use thumbprint) Date

_____ _____

Signature (or thumbprint) of Participant Time

_____ _____

Name of Staff Obtaining Consent Signature & Date

_____ _____ _____

Name of Witness (*if required)* Signature & Date

 IRB No. _____

Statement of Consent to be audiotaped.

I understand that audio recordings will be taken during the study. *(Please choose YES or NO by inserting your initials in the relevant box)*

- I agree to **be audio recorded** **Yes** ☐

No ☐

_____ _____ _____

Name of Participant (please print) Signature Date

YOU WILL BE OFFERED A COPY OF THIS CONSENT FORM TO KEEP.

If you have any questions concerning this study or consent form beyond those answered by the investigator, including questions about the research, your rights as a research participant or research-related injuries; or if you feel that you have been treated unfairly and would like to talk to someone other than a member of the research team, please feel free to contact the Medical Research Council of Zimbabwe (MRCZ) on telephone (04)791792 or (04) 791193 and cell phone lines 0784 956 128. The MRCZ Offices are located at the National Institute of Health Research premises at Corner Josiah Tongogara and Mazowe Avenue in Harare.

11.7.3 Caregivers consent from - Phase 1 (Shona version)

 IRB No. _____

School of Health and Rehabilitation Sciences

Faculty of Health Sciences

Divisions of Communications Sciences and Disorders, Disability Studies, Nursing and Midwifery, Occupational Therapy, Physiotherapy

F45 Old Main Building, Groote Schuur Hospital,

Observatory 7925

Tel: +27 (0) 21 406 6401 Fax: +27 (0) 21 406 6323

Internet: www.uct.ac.za

Zita retsvakiridzo: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Muongorori: Dambi Jermaine, [Msc. PT, Bsc. (Hons) PT]

Varairidzi vangu: Professor J Jelsma, Mrs T Mlambo

Numba dzangu: 0773 444 911

Zvamunofanirwa kuziva pamusoro petsvakiridzo iyi:

- Takupayi hwaro iyi kuti muzive donhodzo yetsvakiridzo iyi.
- Kurapwa muzvipatara kune donhodzo rokuti varwere vapore. Kurapwa uku kunotouya mushure mevongororo yekunyatsoona nzira dzakanyatsotwasanuka dzokurapa nadzo. Donhodzo yetsavaridzo ndoyekuburitsa nzira idzi dzokuti vanhu varapwe zvineruzivo nehuchenjeri.
- Hativimbise kuti tsvakiridzo iyi ichakubatsirayi imi nomwana venyu.
- Munekodzero yekuti mupinde kana kuramba kupinda mutsvakiridzo iyi.
- Munogona kuchinja sarudzo yenyu pasina anokupokana nesarudzo yenyu.
- Zvamunenge masarudza hazvizotadzisi imi nemwana kuti murapwe zvakanaka muneramangwana.
- Nyatsoverengayi hwaro iyi uyezve sukungukayi kubvunza mibvunzo musati maita sarudzo yokupinda mutsvakiridzo.

ZVAMUNOFANIRWA KUZIVA PAERERANO NETSVAKIRIDZO IYI? Kune nzira dzakasiyana dzinokwanisa kushandiswa kurapa nadzo vana vakaremara. Somuenzaniso, vana vanogona kuuya kuchipatara vachibva kumba kwavo kuti vazorapwe mushure mazvo vodzokera zvavo kumba. Zvimwe zvipatara ndizvo zvinedzimba dzokuti vana nevabereki vavo vagarire pachipatara panguva apo vana vachinge vachirapwa ayo anonzi ma rehabilitation villages. Hazvizivikanwi kuti ndeipi nzira inonyatsogutsa vabereki uyezve inonyatsoti vana vanyatsorapwe zvinemazvo. Izvi zvakakosha nokuti zvakavongororwa muzvidzidzo kuti vanhu vanochengeta vana vakaremara vanovanzosangana nematambudziko akasiyana-siyana. Naizvozvo, takagadzira hwaro inemibvunzo yekuti tinzwisise mamwe ematambudziko amunosangana navo mukuchengeta vana vakaremara. Tikaziva matambudziko aya, zvinogona kutibatsira kuti tivane nzira dzokuti tikubatsireyi kurerutsa mutoro wekuchengeta mwana akaremara. Muri kukumbirwa kuti mupinde mutsvakiridzo iyi sezvo muchichengeta/mune mwana ane cerebral palsy/akaremara.

ZVINOTARISIRWA KWAMURI? Kana mukabvuma kupinda mutsvakiridzo iyi, tichakubvunzai mibvunzo inoenderana nekuchengeta mwana akaremara. Tichatapa mhinduro dzamuchatipa uyezve tinotasira kuti kukubvunzai mibvunzo kunogona kutora nguva inosvika mamititsi makumi matatu zvichidzika. Nguva yacho ichaenderana nemhinduro dzamuchange muchipa. Mushure mazvo, tichazokukumbirayi kuti muzopindure mibvunzo yakanyorwa pasi maererano nematambudziko anosangana nevanhu vanochengeta vana vakaremara. Tinotarisira kuti danho iri retsvagiridzi kuti rinogona kutora masvondo maviri kuti ripere. Zvisineyi, tinotarisira kuti munogona kupindura mibvunzo yose muzuva rimwe chete. Tichazenge tichikumbirayi mvumo pose patinenge tichida kukubvunzayi mibvunzo.

PANE NJODZI HERE DZAMUNGATARISIRE? Hatitarisire kuti mungasangana nenjodzi kana mukasarudza kupinda mutsvakiridzo iyi. Zvisineyi tinotarisira kukutorerayi nguva yenyu shomanane apo tichange tichikubvunzayi mibvunzo.

PANE MUBAYIRO HERE WEKUPINDA MUTSVAKIRIDZO? Hapana mibayiro ichapihwa kune avo vachazvipira kupinda mutsvagiridzo iyi. Zvisinei, kana mapinda mutsvikiridzo, tichakupayi imi nemwana zvekudya nekunwa mushure mekunge mapedza kupindura mibvunzo. Pamusoro pazvo, kana muchida rumwe rubatsiro rwurimaererano netsvakiridzo iyi, ndinogona kukubatsirayi nepandinogona kana kuti ndinokuudzayi vanhu kana tsvimbo yamunogona kuvana rubatsiro.

PANE ANOKWANISA HERE KUZIVA ZVAMUCHATIUDZA? Tichachengetedza nekutsindidza zvose zvamuchatiudza. Hatisikuzoshandisa mazita kuitira kuti vanhu vasakwanise kuzoziva zvamunenge mataura. Tichatapa mhinduro dzenyu pama DVD ayo tichakiriya muhofisi iri kuUniversity of Zimbabwe. Ndichachengetadza svumburuno yacho kuti parege kuva nemunhu anokwanisa kuona ma DVD aya. Pamusoro pazvo, maDVD aya tichazomapisa kana gore rapfuura kubva musi watichapedza tsvakiridzo iyi. Tichachengetera mhinduro dzose dzamuchatipa kuUniversity of Zimbabwe, papepa ose kachakiyirwa muhofisi yangu. Tichakokuunganidzayi panzvimbo imwe chete apo tichazokupakurirayi zvinenge zvabuda mutsvakiridzo iyi. Tichazvinyora pasi zvakare kuti muzondogoverana nevamwe zvinenenge zvabuda mutsvakiridzo.

PANE IMWE MARI HERE KAMUNOFANIRWA KUZOSHANDISA PANGUVA YETSVAKIRIDZO?
Hamutarisirwe kunzi muchazoshandisazve imwe mari kuti mupinde

mutsvakiridzo sezvo tichange tichikubvunzayi mibvunzo apo munouya kuzorapisa vana venyu samazuva ose.

MUNOSUNGIRWA HERE KUPINDA MUTSVAKIRIDZO? Hamusungirwi kuti mupinde mutsvakiridzo iyi. Tinokwanisa kukubvunzayi mibvunzo mushure mekunge imi matotipa mvumo. Makasununguka kuramba kupinda mutsvakiridzo. Kuramba kukwikwidza hakukanganise kurapwa kwemwana muneramangwana. Makasununguka kubvunza mibvunzo yose yamunogona kunge muinayo musati maita mhereredzo yekuti mupinde here kana kuti kwete. Makasununguka zvakare kuzochinja mheredzo yenyu chero pamunenge mada pasina kupa tsananguro. Asi kana muchinge mafunga kuzoregedza zvetsvakiridzo, ndinokumbiravo kuti muzondizivisevo.

 IRB No. _____

SIGNATURE PAGE

Musoro wetsvakiridzo: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Protocol Version Number/date

KO KANA MUINE MIBVUNZO? Musazeze zvenyu kundibvunza mibvunzo kana pane zvamunoda kunyatsonzwisisa pamusoro petsvakiridzo iyi. Sunungukayi zvenyu kutora nguva yenyu muchinyatsozeya kuti mopinda mutsvakiridzo here kana kuti kwete.

MVUMO YENYU

Makuita sarudzo yekuti mupinde mutsvakiridzo iyi kana kuti kwete. Mukasayina zvinoreva kuti maverenga hwaro iyi mukainzwisisa, kuti mibvunzo yenyu yose yapindurwa uyezve kuti masarudza kupinda mutsvakiridzo iyi.

_____ _____

Zita renyu (Nyorari rose kana kudhinda) Zuva

_____ _____

Siginecha yesu (Munogona kudhinda) Nguva

_____ _____

Zita remuongorori Siginecha nezuva

_____ _____ _____

Hwitinesi Siginecha nezuva

Mvumo yekuti mutapwe.

Ndinonzwisisa kuti zvandichataura patsvakiridzo iyi zvichatapwa (Ndinokumbira kuti munyore zita renyu mubhokisi rakanzi hongu kana kwete zvichienderana nesarudzo yenyu)

- Ndinobvuma kutapwa **Hongu** ☐

Kwete ☐

_____ _____ _____

Zita renyu (nyorayi kana kudhinda) Siginecha Zuva

MUCHAPIHWA HWARO RENYU RAMUCHAENDA NARO.

Kana muine mubvunzo kana kugunun'una pamusoro petsvakiridzo iyi, sunungukayi zvenyu kuti mubate vanamazvikokota vanoona nezvetsvakiridzo munyika ino vanova veMedical Research Council of Zimbabwe (MRCZ). Nhamba dzavo dzorungare dzinoti (04)791792 kana (04) 791193 uyezve munogona kuvabata pambozha runhare inoti 0784 956 128. VeMRCZ vanovanikwa paNational Institute of Health Research iyo iri panaJosiah Tongogara naMazowe Avenue muHarare.

11.7.4 Caregivers consent form – Phase 2 (English version)

IRB No. _____

School of Health and Rehabilitation Sciences

Faculty of Health Sciences

Divisions of Communications Sciences and Disorders, Disability Studies, Nursing and Midwifery, Occupational Therapy, Physiotherapy

F45 Old Main Building, Groote Schuur Hospital,

Observatory 7925

Tel: +27 (0) 21 406 6401 Fax: +27 (0) 21 406 6323

Internet: www.uct.ac.za

Study title: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Principal Investigator: Dambi Jermaine, [Msc. PT, Bsc. (Hons) PT]

Co-Investigator(s): Professor J Jelsma, Mrs T Mlambo

Phone number(s): 0773 444 911

What you should know about this research study:

- We give you this consent so that you may read about the purpose, risks, and benefits of this research study.
- Routine care is based upon the best-known treatment and is provided with the main goal of helping the individual patient. The main goal of research studies is to gain knowledge that may help future patients.
- We cannot promise that this research will benefit your child. Just like regular care, this research can have side effects that can be serious or minor.
- You have the right to refuse to allow your child to take part or agree for your child to take part now and change your mind later.
- Whatever you decide, it will not affect your child's regular care.
- Please review this consent form carefully. Ask any questions before you make a decision.
- You are free not to answer any questions
- Your choice to allow your child to participate is voluntary.

PURPOSE

They are different ways of providing rehabilitation services. One method is whereby clients walk into a hospital for rehabilitation services straight from their homes. However, some hospitals offer services through rehabilitation villages. These are housing structures whereby caregivers and children with disabilities camp at the district hospitals for up to three days. Their children get rehabilitation treatment during the stay. However, nothing is known about the best method of delivering rehabilitation services. This study hopes to find the best way of providing rehabilitation services as well as ways of providing support for caregivers. This is because people who care for children with disabilities may face certain challenges in looking after these children. It is against this background that we have developed a questionnaire to try to understand your experiences in taking of a child with cerebral palsy (CP). Thus, the findings of this research will be used to develop ways to assist caregivers in coping with the challenges faced in providing care to a child with a disability. Since you have a child with CP, we would like to invite you to take part in our study.

PROCEDURES AND DURATION

If you decide to participate, we will request you to fill in five questionnaires based on your experiences in providing for care to a child with a disability. As the questionnaires are brief, we anticipate that it will take between 30 to 45 minutes to fill in the questionnaires at most. This phase of the study is expected to run for two to four weeks. You will be required to sign a consent form whenever you will be asked to fill in questionnaires.

RISKS AND DISCOMFORTS

This study will not pose any danger to you except that we may require some of your time which may disrupt your daily schedule on the day we will ask you questions.

BENEFITS AND/OR COMPENSATION

There are no associated benefits in participating in this study. However, you (and the children) will be provided with food and drinks after the interviews and upon completion of the questionnaires. Additionally, should require any further treatment and support, the researcher will be glad to assist you and or refer you to other people who can assist you.

CONFIDENTIALITY

Information gathered about you, and your child will be kept in private. You will be assigned a code/number for identification purposes, and no names will be used for reference in the study and publications. In addition, only the researcher and under some circumstances, the MRCZ may need to review patient records for compliance audits. We will inform you about the outcomes of the present study through oral presentations and leaflets with simplified and summarized information at the end of the study.

ADDITIONAL COSTS

There are no additional costs associated with your participation in this study as we will ask you questions on the days you bring your child for the usual treatments.

VOLUNTARY PARTICIPATION

Participation in this study is voluntary. If you decide not to participate in this study, your decision will not affect your future relations with the hospital and its personnel. If you decide to participate, you are free to withdraw your consent and to discontinue participation at any time without penalty. However, in the event that you choose to withdraw, please do try by all means to notify the researcher about your decision.

 IRB No. _____

SIGNATURE PAGE

Study title: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Protocol Version Number/date

OFFER TO ANSWER QUESTIONS

Before you sign this form, please ask any questions on any aspect of this study that is unclear to you. You may take as much time as necessary to think it over.

AUTHORIZATION

You are making a decision whether or not to participate in this study. Your signature indicates that you have read and understood the information provided above, have had all your questions answered, and have decided to participate.

_____ _____

Name of Research Participant (please print/ use thumbprint) Date

_____ _____

Signature (or thumbprint) of Participant Time

_____ _____ _____

Name of Staff Obtaining Consent Signature & Date

_____ _____ _____

Name of Witness (*if required)* Signature & Date

YOU WILL BE OFFERED A COPY OF THIS CONSENT FORM TO KEEP.

If you have any questions concerning this study or consent form beyond those answered by the investigator, including questions about the research, your rights as a research participant or research-related injuries; or if you feel that you have been treated unfairly and would like to talk to someone other than a member of the research team, please feel free to contact the Medical Research Council of Zimbabwe (MRCZ) on telephone (04)791792 or (04) 791193 and cell phone lines 0784 956 128. The MRCZ Offices are located at the National Institute of Health Research premises at Corner Josiah Tongogara and Mazowe Avenue in Harare.

11.7.5 Caregivers consent form – Phase 2 (Shona version)

IRB No. _____

School of Health and Rehabilitation Sciences

Faculty of Health Sciences

Divisions of Communications Sciences and Disorders, Disability Studies, Nursing and Midwifery, Occupational Therapy, Physiotherapy

F45 Old Main Building, Groote Schuur Hospital,

Observatory 7925

Tel: +27 (0) 21 406 6401 Fax: +27 (0) 21 406 6323

Internet: www.uct.ac.za

Zita retsvakiridzo: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Muongorori: Dambi Jermaine, [Msc. PT, Bsc. (Hons) PT]

Varairidzi vangu: Professor J Jelsma, Mrs T Mlambo

Numba dzangu: 0773 444 911

Zvamunofanirwa kuziva pamusoro petsvakiridzo iyi:

- Takupayi hwaro iyi kuti muzive donhodzo yetsvakiridzo iyi.
- Kurapwa muzvipatara kune donhodzo rokuti varwere vapore. Kurapwa uku kunotouya mushure mevongororo yekunyatsoona nzira dzakanyatsotwasanuka dzokurapa nadzo. Donhodzo yetsavaridzo ndoyekuburitsa nzira idzi dzokuti vanhu varapwe zvineruzivo nehuchenjeri.
- Hativimbise kuti tsvakiridzo iyi ichakubatsirayi imi nomwana venyu.
- Munekodzero yekuti mupinde kana kuramba kupinda mutsvakiridzo iyi.
- Munogona kuchinja sarudzo yenyu pasina anokupokana nesarudzo yenyu.
- Zvamunenge masarudza hazvizotadzisi imi nemwana kuti murapwe zvakanaka muneramangwana.
- Nyatsoverengayi hwaro iyi uyezve sukungukayi kubvunza mibvunzo musati maita sarudzo yokupinda mutsvakiridzo.

ZVAMUNOFANIRWA KUZIVA PAERERANO NETSVAKIRIDZO IYI? Kune nzira dzakasiyana dzinokwanisa kushandiswa kurapa nadzo vana vakaremara. Somuenzaniso, vana vanogona kuuya kuchipatara vachibva kumba kwavo kuti vazorapwe mushure mazvo vodzokera zvavo kumba. Zvimwe zvipatara ndizvo zvinedzimba dzokuti vana nevabereki vavo vagarire pachipatara panguva apo vana vachinge vachirapwa ayo anonzi ma rehabilitation villages. Hazvizivikanwi kuti ndeipi nzira inonyatsogutsa vabereki uyezve inonyatsoti vana vanyatsorapwe zvinemazvo. Izvi zvakakosha nokuti zvakavongororwa muzvidzidzo kuti vanhu vanochengeta vana vakaremara vanovanzosangana nematambudziko akasiyana-siyana. Naizvozvo, takagadzira hwaro inemibvunzo yekuti tinzwisise mamwe ematambudziko amunosangana navo mukuchengeta vana vakaremara. Tikaziva matambudziko aya, zvinogona kutibatsira kuti tivane nzira dzokuti tikubatsireyi kurerutsa mutoro wekuchengeta mwana akaremara. Muri kukumbirwa kuti mupinde mutsvakiridzo iyi sezvo muchichengeta/mune mwana ane cerebral palsy/akaremara.

<u>ZVINOTARISIRWA KWAMURI?</u> Kana mukabvuma kupinda mutsvakiridzo iyi, ndichakukumbirayi kuti mupindure mibvunzo iri pamapepa mashanu inoenderana nematambudziko amunosangana navo zuva-nezuva mukuchengeta vana. Sezvo mibvunzo yacho irimishoma, tinotarisira kuti zvinogona kutora nguva inokwana maminitsi makumi matatu. Tinotarisira kwakare kuti danho iri retsvakiridzo kuti rinogona kutora masvondo mana kuti ripere. Zvisineyi, tinotarisira kuti munogona kupindura mibvunzo yose muzuva rimwe chete. Tichazenge tichikukumbirayi mvumo pose patinenge tichida kukubvunzayi mibvunzo.

PANE NJODZI HERE DZAMUNGATARISIRE? Hatitarisire kuti mungasangana nenjodzi kana mukasarudza kupinda mutsvakiridzo iyi. Zvisineyi tinotarisira kukutorerayi nguva yenyu shomanane apo tichange tichikubvunzayi mibvunzo.

PANE MUBAYIRO HERE WEKUPINDA MUTSVAKIRIDZO? Hapana mibayiro ichapihwa kune avo vachazvipira kupinda mutsvagiridzo iyi. Zvisinei, kana mapinda mutsvikiridzo, tichakupayi imi nemwana zvekudya nekunwa mushure mekunge mapedza kupindura mibvunzo. Pamusoro pazvo, kana muchida rumwe rubatsiro rwurimaererano netsvakiridzo iyi, ndinogona kukubatsirayi nepandinogona kana kuti ndinokuudzayi vanhu kana tsvimbo yamunogona kuvana rubatsiro.

PANE ANOKWANISA HERE KUZIVA ZVAMUCHATIUDZA? Tichachengetedza nekutsindidza zvose zvamuchatiudza. Hatisikuzoshandisa mazita kuitira kuti vanhu vasakwanise kuzoziva zvamunenge mataura. Tichachengetera mhinduro dzose dzamuchatipa kuUniversity of Zimbabwe, papepa ose kachakiyirwa muhofisi yangu. Tichakokuunganidzayi panzvimbo imwe chete apo tichazokupakurirayi zvinenge zvabuda mutsvakiridzo iyi. Tichazvinyora pasi zvakare kuti muzondogoverana nevamwe zvinenenge zvabuda mutsvakiridzo.

PANE IMWE MARI HERE KAMUNOFANIRWA KUZOSHANDISA PANGUVA YETSVAKIRIDZO? Hamutarisirwe kunzi muchazoshandisazve imwe mari kuti mupinde mutsvakiridzo sezvo tichange tichikubvunzayi mibvunzo apo munouya kuzorapisa vana venyu samazuva ose.

MUNOSUNGIRWA HERE KUPINDA MUTSVAKIRIDZO? Hamusungirwi kuti mupinde mutsvakiridzo iyi. Tinokwanisa kukubvunzayi mibvunzo mushure mekunge imi matotipa mvumo. Makasununguka kuramba kupinda mutsvakiridzo. Kuramba kukwikwidza hakukanganise kurapwa kwemwana muneramangwana. Makasununguka kubvunza mibvunzo yose yamunogona kunge muinayo musati maita mhereredzo yekuti mupinde here kana kuti kwete. Makasununguka zvakare kuzochinja mheredzo yenyu chero pamunenge mada pasina kupa tsananguro. Asi kana muchinge mafunga kuzoregedza zvetsvakiridzo, ndinokumbiravo kuti muzondizivisevo.

SIGNATURE PAGE

Musoro vetsvakiridzo: Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy

Protocol Version Number/date

KO KANA MUINE MIBVUNZO? Musazeze zvenyu kundibvunza mibvunzo kana pane zvamunoda kunyatsonzwisisa pamusoro petsvakiridzo iyi. Sunungukayi zvenyu kutora nguva yenyu muchinyatsozeya kuti mopinda mutsvakiridzo here kana kuti kwete.

MVUMO YENYU

Makuita sarudzo yekuti mupinde mutsvakiridzo iyi kana kuti kwete. Mukasayina zvinoreva kuti maverenga hwaro iyi mukainzwisisa, kuti mibvunzo yenyu yose yapindurwa uyezve kuti masarudza kupinda mutsvakiridzo iyi.

_____ _____

Zita renyu (Nyorari rose kana kudhinda) Zuva

_____ _____

Siginecha yesu (Munogona kudhinda) Nguva

_____ _____

Zita remuongorori Siginecha nezuva

_____ _____ _____

Hwitinesi Siginecha nezuva

MUCHAPIHWA HWARO RENYU RAMUCHAENDA NARO.

Kana muine mubvunzo kana kugunun'una pamusoro petsvakiridzo iyi, sunungukayi zvenyu kuti mubate vanamazvikokota vanoona nezvetsvakiridzo munyika ino vanova veMedical Research Council of Zimbabwe (MRCZ). Nhamba dzavo dzorungare dzinoti (04)791792 kana (04) 791193 uyezve munogona kuvabata pambozha runhare inoti 0784 956 128. VeMRCZ vanovanikwa paNational Institute of Health Research iyo iri panaJosiah Tongogara naMazowe Avenue muHarare.

11.8 Appendix 8: Harare Central Hospital Central Ethics Committee approval letter

Telephone: 621100-19
Fax: 621157

ZIMBABWE

Reference: HCHEC 110316/18

HARARE CENTRAL HOSPITAL
P. O. Box ST 14
SOUTHERTON
Harare

31 March 2016

Mr. J. Dambi
Department of Rehabilitation
University of Zimbabwe
HARARE

Dear Mr. Dambi,

REF: "DETERMINATION OF THE IMPACT OF REHABILITATION VILLAGES AS A SERVICE DELIVERY MODEL: A STUDY OF VIEWS AND EXPERIENCES OF ZIMBABWEAN CAREGIVERS OF CHILDREN WITH CEREBRAL PALSY"

I am glad to advise you that your application to conduct a study entitled: **"Determination of the Impact of Rehabilitation Villages as a Service Delivery Model: A Study of Views and Experiences of Zimbabwean Caregivers of Children With Cerebral Palsy" (Ref: HCHEC 110316/18),** has been approved by the Harare Hospital Ethics Committee.

This approval is premised on the submitted protocol. Should you decide to vary your protocol in any material way please submit these for further approval.

You are advised to avail the results of your study whether positive or negative to the hospital through the committee for our information.

Yours sincerely,

HARARE CENTRAL HOSPITAL
DEPARTMENT OF MEDICINE
31 MAR 2016
P.O. BOX ST 14, SOUTHERTON
HARARE

DR. C. Pasi
Chairman Harare Central Hospital Ethics Committee

11.9 Appendix 9: Mashonaland West Provisional Medical director approval letter

Reference:

Telephone: 23211-6
Telegraphic Address:
"PROVMED" Chinhoyi
Fax: 23218

E-mail: pmdmashwest@gmail.com

ZIMBABWE

MINISTRY OF HEALTH AND CHILD CARE
PROVINCIAL MEDICAL DIRECTOR
(Mashonaland West Province)
P.O Box 139
Chinhoyi
Zimbabwe

13 April 2016

TO WHOM IT MAY CONCERN

RE: REQUEST FOR PERMISSION TO CARRYOUT PhD STUDY

This serves to confirm that Jermaine M. Dambi studying for a PHD in Phsiotherapy at University of Cape Town, has been granted permission to carryout a research in Mashonaland West Province.

Thank you.

MINISTRY OF HEALTH & CHILD CARE
PROVINCIAL MEDICAL DIRECTOR
MASHONALAND WEST PROVINCE
13 APR 2016
P.O. BOX 139, CHINHOYI
ZIMBABWE TEL: 067-23218

Dr W. Nyamayaro
PROVINCIAL MEDICAL DIRECTOR MASHONALAND WEST PROVINCE

11.10 Appendix 10: Mashonaland Central Provisional Medical director approval letter

Telephone Nos.
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P.M.D. Staff Offices 6369, 6381
6479, 6659
Fax +263 71 6956
P.N.O. 6366
P.H.S.A. 6205.
P.E.H.O. 6204

Reference

MINISTRY OF HEALTH AND CHILD CARE
PROVINCIAL MEDICAL DIRECTOR
Mashonaland Central
P O Box 98
Bindura
Zimbabwe

7 March 2016

The DMOs
Mashonaland Central Province

TO WHOM IT MAY CONCERN

The bearer, Jermaine M. Dambi is a PhD Physiotherapy student at the University of Cape Town (UCT) in South Africa who wishes to conduct a study entitled "**Determination of the Impact of Rehabilitation Villages as a service of delivery : A Study of views and experiences of Zimbabwe Caregivers of children with cerebral Palsy".**

He has been granted permission to undertake the study by the Provincial Medical Director of Mashonaland Central, on condition that he receives ethical clearance from the Medical Research Council of Zimbabwe.

Kindly assist him wherever possible.

Sincerely

MINISTRY OF HEALTH
PROVINCIAL MEDICAL DIRECTOR
MASHONALAND CENTRAL
- 7 MAR 2016
P.O BOX 98
BINDURA
ZIMBABWE

MEDICAL SUPERINTENDENT
BINDURA PROVINCIAL HOSPITAL
07 MAR 2016
P. BAG 940, BINDURA
ZIMBABWE

PERMISSION GRANTED
7.03.16

Dr C Tshuma
PROVINCIAL MEDICAL DIRECTOR – MASHONALAND CENTRAL

Permission Granted Above Conditions Are Met.

DISTRICT MEDICAL OFFICER
MT DARWIN DISTRICT HOSPITAL
HUMAN RESOURCES OFFICER
07 MAR 2016
P.O. BOX 97, MT. DARWIN
ZIMBABWE

11.11 Appendix 11: Director of Health Services City of Harare approval letter

CITY OF HARARE

Director of Health Services

DR PROSPER CHONZI
MBChB, MPH,MBA

All correspondence to be addressed to the
DIRECTOR OF HEALTH SERVICES

Ref:----------

Your Ref:----------

DIRECTOR OF HEALTH SERVICES
Rowan Martin Building,
Civic Centre,
Pennefather Avenue,
off Rotten Row,
Harare, Zimbabwe.

P.O. Box 596
Telephone: 753326
753330/1/2
Fax: (263-4) 752093

03 May 2016

Mr Jermaine M Dambi
University of Zimbabwe
Department of Rehabilitation
P O Box A178
Avondale
HARARE

Dear Madam

RE: REQUEST TO CONDUCT A STUDY

I acknowledge receipt of your letter in connection with the above.

Permission is granted for you to carry out a study entitled "***Determination of the impact of rehabilitation villages as a service delivery model: A study of views and experiences of Zimbabwean caregivers of children with cerebral palsy.***" at Highfield, Kuwadzana, Mabvuku, Mbare, Kuwadzana, Kambuzuma and Mufakose Polyclinics

For further assistance please liaise with the Sisters In Charge of the respective clinics.

Yours faithfully

DIRECTOR OF HEALTH SERVICES
IM/rm

c.c. Ethics committee

S.I.C.	-	Highfiled Polyclinic
S.I.C.	-	Glenview Polyclinic
S.I.C.	-	Mabvuku Polyclinic
S.I.C.	-	Mbare Plyclinic
S.I.C.	-	Kuwadzana Polyclinic
S.I.C.	-	Kambuzuma Polyclinic
S.I.C.	-	Mufakose Polyclinic

11.12 Appendix 12: Research Assistants Training for taking adult consent

Aim - The broad objective is to standardize the data collection procedures to increase the reliability and validity of the study findings

Specific goal - To train the research assistants to obtain consent from caregivers and assent from children accurately were appropriate

Methodology for training for obtaining consent

The research assistants will be trained to obtain consent using a five-step process as illustrated in Figure 2 below:

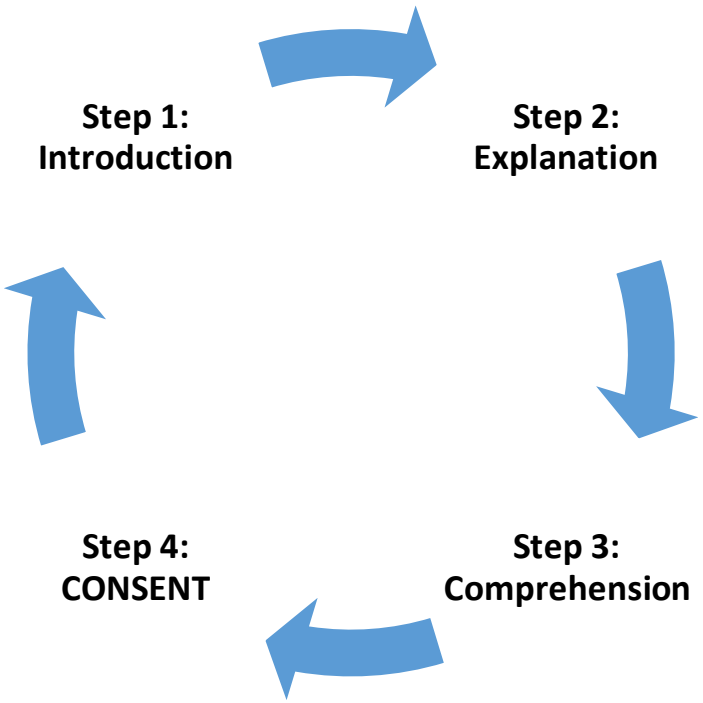

Figure 11-1: Consent process

Introduction

- Greet the prospective participants and introduce themselves
- To avoid cohesion and for the preservation of confidentiality - invite the prospective participants in a private room

Explanation

- Enquire from the participants their preferred language, i.e. between English and Shona
- Explain the study aims and objectives. Emphasis should be on the use of simple and non-technical language
- Offer the prospective participant the written information letter

- Enquire from the prospective participant the preferred mode of administration of the information letter, i.e. whether they prefer to read through the letter themselves or that the research assistant to read to them the information letter
- Give the participant time to go through the information letter and reflect upon the information provided

Comprehension

- Allow prospective participants to ask any questions
- Make use of open-ended/teach back questions to ensure that the prospective participant fully comprehends the information outlined in the information letter before they can make an informed decision. You may ask the following: I want to be sure if you have fully understood the contents of the information letter and or explanations are given. Can you please explain to me the following?

1. *Why are you being requested to take part in the study?*
2. *What will be expected of you if you agree to participate?*
3. *Does it matter if you choose not to take part in the study?*
4. *Are there any benefits associated with participating in the study? "*

- Were appropriate give the prospective feedback so that they may make an informed choice

11.13 Appendix 13: Translation process and adaptation of the MSPSS

11.13.1 Study setting

The study was done at the Children's Rehabilitation Unit (CRU) [12] and rural district hospitals in Zimbabwe.

11.13.2 Sampling

11.13.2.1 Selection criterion

We selected primary caregivers who were; unpaid for the caregiving role, lived with the child [540] and could understand both written and spoken English and Shona. We excluded caregivers if; they were not fluent in Shona and had a confirmed psychiatric diagnosis according to doctors' notes.

11.13.2.2 Sample size calculation

We conveniently recruited 16 caregivers for pilot testing of the MSPSS-Shona version. The sample was derived from the recommendations by the ISPOR guidelines [466].

11.13.3 Instrumentation

11.13.3.1 The original version of the MSPSS

The MSPSS contains 12 items that measure the perceived adequacy of the available amount of SS from three sources, i.e. friends, family and significant other/special person [559,560]. Responses are rated on a seven-point Likert scale; with responses ranging from very strongly disagree (=1) to very strongly agree (=7). The cumulative/total scores range from 12 to 84. As there are no cut-off points, scores are interpreted as; the higher the score, the more significant the amount of available SS [559]. The original version of the MSPSS yielded a three-factor structure, high internal consistency (α=0.88), stability (yielded α=0.85 after three months from initial administration) and moderate construct validity as the SS scores were negatively correlated with anxiety (r=-0.18; p<0.01) and depression scores (r=-24; p<0.01) [560].

11.13.3.2 Self-designed questionnaires

These were developed to measure caregivers' satisfaction with the presentation, clarity of instructions and items and cultural relevancy. Satisfaction was rated on a five-point Likert scale with responses ranging from strongly disagree=1 to strongly agree=5.

11.13.4 Translation procedure

As illustrated in Figure 11-2 below, we utilized a Multiple-stage design adapted from the guidelines by the ISPOR- Translation and Cultural Adaptation Group guidelines [466].

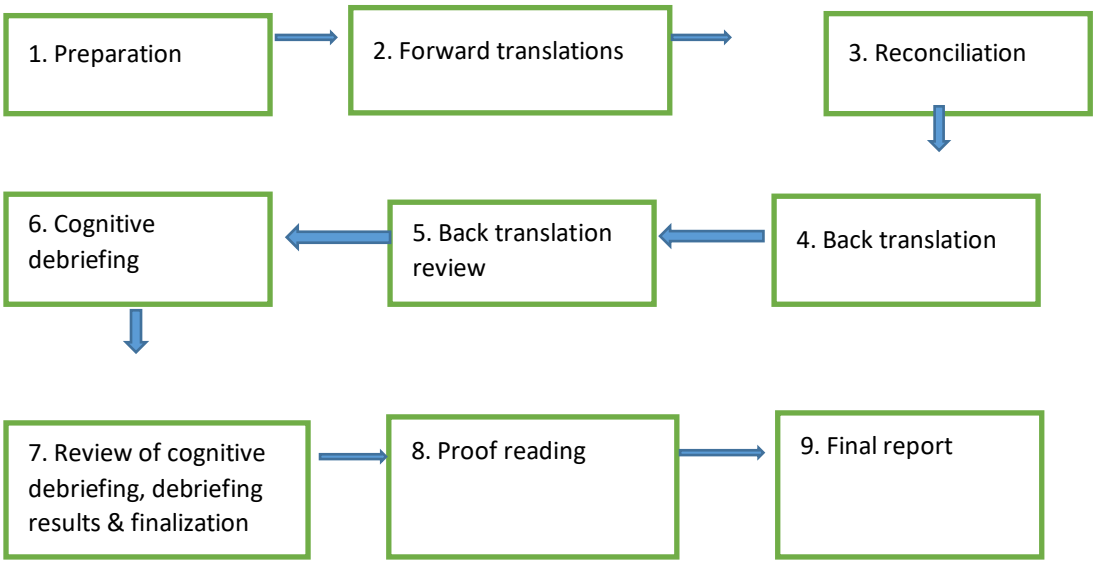

Figure 11-2: Translation process of the MSPSS

11.13.4.1 Step one: preparation

The developer granted us permission to adapt and translate the MSPSS. The developer was also invited to be involved in the translation process if clarification of any conceptual ambiguities was needed. Lastly, we recruited personnel who aided in the translation process.

11.13.4.2 Step two: forward translation

Two translators blindly translated the MSPSS into Shona. The emphasis was on attaining a colloquial translation. The translators were a Linguistics professor (T1) and a senior Linguistics lecturer (T2) from the African Languages Research Institute and University of Zimbabwe Linguistics Department respectively. T1 was not familiar with the concepts enshrined within the MSPSS and had no prior experience in translating PROMs; this was essential to get a literal translation. T2 had prior experience in translating PROMs; this was important to ensure the attainment of a more conceptually equivalent translation. Both translators are prolific English and Shona speakers and worked independently.

11.13.4.3 Step three: reconciliation

An independent translator (T3) who was not involved in the forward translation reconciled the two forward translations into one version. T3 is a Senior Shona lecturer in the Department of Linguistics at Harare Polytechnic College. After that, there was a discussion between the two forward translators (T1 & T2), the PI, reconciliation translator (T3), and co-investigator to harmonise the reconciled Shona version.

11.13.4.4 Step four: back translation

The Shona version was then translated back to English by two independent translators (T4 & T5) who are fluent Shona and English speakers. The emphasis of the translation was in literal and conceptual meaning. The translators were senior lecturers in the Departments of English and Linguistics at the University of Zimbabwe, and both were not involved in the forward translation process. Only translator T4 had prior experience and expertise in PROs translations.

11.13.4.5 Step five: back translation review

The backward translation was compared against the original MSPSS *to ensure the conceptual equivalence of the translation* [466]*,* the PI did this with the help of the MSPSS developer.

11.13.4.6 Step six: cognitive debriefing

The translated tool was then administered to a group of caregivers of children with CP, n=16. Initially, the Shona questionnaire was administered after which the caregivers were requested to complete the English version. Respondents also evaluated the clarity of instructions, cultural relevancy, and the layout of the questionnaire.

11.13.4.7 Step seven: a review of cognitive debriefing and finalisation

This was done by the principal researcher with the assistance from the co-investigators. This step was essential in ensuring cultural relevance by finding items which could have required modification or rewording.

11.13.4.8 Step eight: proofreading

This was done to check for any typo errors which could have been missed during the translation process. This was done by an independent linguist and a physiotherapist experienced in translation of outcome measurements who were not involved in the initial translation process to eliminate possibilities of any bias.

11.13.4.9 Step nine: final report

This step involved a write up of the methodology utilized and critical decisions/amendments made during the translation process.

11.13.4.10 The rationale for study design

The developers of the MSPSS did not lay down the guidelines for the translation of their tool thus the choice of the ISPOR guidelines. The ISPOR guidelines were deemed appropriate as they were built upon the review and consultation of 12 other standardized translation and adaptation guidelines [466].

11.13.5 Data analysis

Differences between translation versions were analysed qualitatively. We also used frequencies and the Spearman correlation coefficient to analyse the relationship between scores on the English and Shona versions of the MSPSS during the cognitive debriefing stage.

11.13.6 Data Management

Raw data were de-identified by coding the data, and files containing the raw data were kept in a locked and secure drawer at the University of Zimbabwe Department of Rehabilitation.

11.13.7 Results

11.13.7.1 Reconciliation of forward translations

Illustrated in Table 11.1 below is the reconciliation process of the two forward translations. As they were few discrepancies across the translations, we adopted a translation which was; brief, simple to understand, more polite/respectful, and succulently captured the underlying concept. We avoided ambiguous words for example, for item 1, we dropped the translation with the term "dambudziko" an equivalent to "problem". The term has several conceptual meanings, i.e. it can mean "need", "trouble", "grief" or "challenge". Further, both translations did not succinctly capture the conceptual equivalency for item 5 thus the amendment through a consensus discussion. For item 10, no exact equivalent for the word "feelings" in the Shona language was found.

Table 11.1 : Reconciliation of MSPSS forward translations

Version	Item	Rationale for decisions/notes
English	Instructions: We are interested in how you feel about the following statements. Read each statement carefully. Indicate how you feel about each statement	
T1	Zvekuita: Tinoda kunzwa zvaunofunga pane zvinotevera. Nyatsoverenga mutsara woga uchiratidza mawonero ako sekutaurwa kwazvakaitwa. Ratidza zvaunofunga nekutara denderedzwa.	
T2	Zvinofanira kutevedzwa: Tinoda kuziva pfungwa dzenyu maererano nezvirevo zvinotevera. Nyatsoverengai chirevo chimwe nechimwe mugotaridza pfungwa dzenyu pachirevo chimwe nechimwe	
Adopted		
English	Circle the "1" if you Strongly Disagree	

T1	Tenderedza “1” kana usingatenderane nazvo zvakanyanya	
T2	Komberedzai “1” kana musingabvumirani nazvo zvachose	
Adopted	Tenderedzai “1” kana musingatenderane nazvo zvakanyanya	
English	Circle the “2” if you Disagree	
T1	tenderedza “2” kana usingatenderane nazvo	
T2	Komberedzai “2” kana musingabvumirani nazvo	
Adopted	tenderedzai “2” kana musingatenderane nazvo	
English	Circle the “3” if you are Neutral	
T1	tenderedza “3” kana usina zvaunotenderana kana kusatenderana nazvo	
T2	Komberedzai “3” kana muri pakati nepakati	
Adopted	Tenderedzai “3” kana muri pakati nepakati	
English	Circle the “4” if you Agree	
T1	tenderedza “4” kana uchitenderana nazvo	
T2	Komberedzai “4” kana muchibvumirana nazvo	
Adopted	Tenderedzal “4” kana muchitenderana nazvo	
English	Circle the “5” if you Strongly Agree	
T1	tenderedza “5” kana uchitenderana nazvo zvakanyanya	
T2	Komberedzai “5” kana muchibvumirana nazvo chose	
Adopted	Tenderedzai “5” kana muchitenderana nazvo zvakanyanya	
English	1. There is a special person who is around when I am in need	Although the first translation was brief, the term “dambudziko” has several conceptual meanings, e.g. it can mean need, trouble, grief, challenge
T1	Pane munhu anokosha anondibatsira kana ndiine dambudziko	
T2	Pane munhu akakosha anomira neni pandinenge ndichida rubatsiro	

Adopted	Pane munhu akakosha anomira neni pandinenge ndichida rubatsiro	
English	2. There is a special person with whom I can share joys and sorrows	• Expression “wandinotaurira” is more respectful than “andinotaurira.” • The expression for the term “There is a special person…” was the same for both translators
T1	Pane munhu anokosha wandino kurukura naye mukufara nemukusuva	
T2	Pane munhu akakosha andinotaurira zvinondifadza nezvinondisuwisa.	
Adopted	Pane munhu anokosha wandinotaurira zvinondifadza nezvinondisuwisa	
English	3. My family really tries to help me	Both translations were almost the same. However, the first translation omitted the expression “really” which was captured as “chose” in the second translation
T1	Vemhuri yangu vanoedza kundibatsira	
T2	Vemhuri yangu vanoedza chose kundibatsira	
Adopted	Vemhuri yangu vanoedza chose kundibatsira	
English	4. I get the emotional help & support I need from my family	• The first translation missed the essence of both “help” and “support” enshrined in the item • Both translations demonstrated that there is no local equivalent/exact translation for the term “emotional.”
T1	Ndinowana rubatsiro kubva kune vemhuri kana pfungwa dzangu dzisina kugadzikana	
T2	Ndinowana rubatsiro nerutsigiro rwandinoda pane zvinondinetsa mupfungwa kubva kumhuri yangu	
Adopted	Ndinowana rubatsiro nerutsigiro rwandinoda pane zvinondinetsa mupfungwa kubva kumhuri yangu	
English	5. I have a special person who is a real source of comfort to me	• Both translations did not succinctly capture the underlying construct thus the amendment during reconciliation • Translation 2 was considered as “too deep” navicular which might be potentially confusing to the younger generation of carers
T1	Pane munhu anokosha anondinyaradza kana ndiine matambudziko	
T2	Ndine munhu akakosha anonyatsondipa rudekaro.	
Adopted	Ndine munhu anokosha anonyatsondinyaradza	
English	6. My friends really try to help me	Both translations were almost the same. However, the first translation did not accurately translate the term “really”, “dzinoedzawo” can mean “slightly” or “somewhat” thus the choice for the second translation
T1	Shamwari dzangu dzinoedzawo kundibatsira	
T2	Shamwari dzangu dzinoedza chose kundibatsira	
Adopted	Shamwari dzangu dzinoedza chose kundibatsira	
English	7. I can count on my friends when things go wrong	• Both translations were almost the same

T1	Ndinovimba ne shamwari dzangu kana zvinhu zvisingafambe zvakanaka	• We opted for the second translation as it was brief and succulent
T2	Ndinovimba neshamwari dzangu kana zvinhu zvikakanganisika	
Adopted	Ndinovimba neshamwari dzangu kana zvinhu zvikakanganisika	
English	8. I can talk about my problems with my family	• Both translations were almost the same • "kutaura" is a more literal translation whereas "kukurukura" accurately captures the concept of discussing/talking about problems
T1	Ndinokwanisa kutaura matambudziko angu nevemhuri yangu.	
T2	Ndinokwanisa kukurukura matambudziko angu nemhuri yangu.	
Adopted	Ndinokwanisa kukurukura matambudziko angu nemhuri yangu.	
English	9. I have friends with whom I can share my joys and sorrows.	• Both translations accurately captured the underlying concepts • Translation 2 was chosen because of its brevity
T1	Ndine shamwari dzandinokwanisa kukurukura nadzo munguva dzekufara nekusuva.	
T2	Ndine shamwari dzandinogona kutaurira zvinondifadza nezvinondisuwisa.	
Adopted	Ndine shamwari dzandinokwanisa kutaurira zvinondifadza nezvinondisuwisa.	
English	10. There is a special person in my life who cares about my feelings	• Both translations accurately captured the underlying concepts. However, there is no exact equivalent for the word "feelings" in the Shona language. • We decided to blend both translations to improve clarity
T1	Pane munhu anokosha muupenyu hwangu ane hanya nezvandinonzwa	
T2	Ndine munhu akakosha muupenyu hwangu anokoshesa mafungiro angu.	
Adopted	Pane munhu anokosha muupenyu hwangu ane hanya nemafungiro nezvandinonzwa	
English	11. My family is willing to help me make decisions.	• Both translations accurately captured the underlying concepts • We opted for the brief translation
T1	Vemhuri yangu vanondibatsira kuronga nezvehupenyu hwangu.	
T2	Mhuri yangu inoda kundibatsira kusarudza zvekuita.	
Adopted	Mhuri yangu inoda kundibatsira kusarudza zvekuita.	
English	12. I can talk about my problems with my friends	Both translations accurately captured the underlying concepts and were the same

T1	Ndinokwanisa kukurukura matambudziko angu neshamwari dzangu	
T2	Ndinokwanisa kukurukura matambudziko angu neshamwari dzangu.	
Adopted	Ndinokwanisa kukurukura matambudziko angu neshamwari dzangu.	

11.13.7.2 Harmonization of backward translations

As illustrated in Table 11.2 below, they were few apparent discrepancies between the backward translations. The emphasis was on getting a brief and succulent translation to decrease respondent burden. Items 2, 4 and 10 were slightly problematic as they were lengthy and contained some jargon and the harmonisation was attained through discussion.

Table 11.2: Harmonization of MSPSS backward translations

T4	Instructions: We would like to know what you think about the following issues. Read carefully every sentence indicating what you feel exactly about each instance. Indicate your choice with a circle		
T5	Activities: We want your opinions about the following. Carefully read every sentence/line and give your opinion as directed in the instructions. Show your opinion/views by encircling the answer of your choice.		
Reconciliation	Instructions: We would like to know what you think about the following issues. Carefully read every sentence and give your opinion as directed in the instructions. Show your opinion/views by encircling the answer of your choice.		
	T4	T5	Reconciliation
	Encircle “1” if you strongly disagree	Encircle “1” if you strongly disagree	Encircle “1” if you strongly disagree.
	encircle “2” if you disagree	Encircle “2” if you disagree.	Encircle “2” if you disagree
	encircle “3” if you are indifferent	Encircle «3» if you are neutral.	Encircle “3” if you are indifferent
	encircle “4” if you agree	Encircle “4” if you agree.	Encircle “4” if you agree.
	encircle “5” if you strongly agree	Encircle “5” if you strongly agree.	Encircle “5” if you strongly agree.
T4	1. There is someone close who assists whenever I face problems		
T5	A very special person helped me in my time of need/ when I had a serious problem.		
Reconciliation	There is a very special person who assists me whenever I face problems		
T4	2. I have someone close with whom I talk to about issues that make me happy or sad		

T5	There is someone special whom I confide in when good things happen to me and when I am also troubled
Reconciliation	There is someone special whom I talk to about issues that make me happy or sad
T4	3. My family try by all means to help me
T5	My family members always try very hard to help me.
Reconciliation	My family try by all means to help me
T4	4. I get assistance and support that I need from my family when I have something troubling my mind
T5	I get help and all the support that I need psychologically from my family.
Reconciliation	I get help and support that I need from my family when I have something troubling my mind
T4	5. There is someone important who consoles me in difficult times
T5	There is a special person who consoles me whenever I am in trouble or have problems.
Reconciliation	There is a special person who consoles me in difficult times
T4	6. My friends try by all means to assist me
T5	My friends always stand by me and give me all the support/help that I need.
Reconciliation	My friends try by all means to assist me
T4	7. I depend on my friends in difficult moments
T5	I trust my friends when things go wrong/when I mess up.
Reconciliation	I depend on my friends when things go wrong
T4	8. I can discuss my problems with my family
T5	I am able to discuss my problems with my family.
Reconciliation	I can discuss my problems with my family
T4	9. I have friends with whom I share issues that make me happy or sad.
T5	I have friends whom I can open up to about my joys and sorrows.
Reconciliation	I have friends with whom I share my joys and sorrows.
T4	10. There is someone close in my life who cares about how I feel
T5	There is a very special person in my life who is deeply concerned about how I feel.
Reconciliation	There is a very special person in my life who cares how I feel.

T4	11. My family offers to help me to make my choices.
T5	My family is ready to help me make choices.
Reconciliation	My family is ready to help me make choices
T4	**12.** I discuss my problems with friends.
T5	I am able to discuss my problems with my friends
Reconciliation	I discuss my problems with friends

11.13.7.3 Backward translation review

The developer confirmed that the backward translation was conceptually equivalent to the original although they were slight idiomatic and grammatic discrepancies. The lack of an exact equivalent for the expression "emotion" resulted in a lengthy backward translation for item 4 (See Table 11.3 below).

Table 11.3: Backward translation review of the MSPSS

Original version	**Instructions: We would like to know what you think about the following issues. Read carefully every sentence indicating what you feel exactly about each instance. Indicate your choice with a circle**	
Forward translation	Instructions: We would like to know what you think about the following issues. Carefully read every sentence and give your opinion as directed in the instructions. Show your opinion/views by encircling the answer of your choice.	
	Original version	Forward translation
	Circle the "1" if you Strongly Disagree	Encircle "1" if you strongly disagree.
	Circle the "2" if you Disagree	Encircle "2" if you disagree
	Circle the "3" if you are Neutral	Encircle "3" if you are indifferent
	Circle the "4" if you Agree	Encircle "4" if you agree.
	Circle the "5" if you Strongly Agree	Encircle "5" if you strongly agree.
Original version	1. There is a special person who is around when I am in need.	
Forward translation	There is a very special person who assists me whenever I face problems	
Original version	2. There is a special person with whom I can share joys and sorrows.	
Forward translation	There is someone special whom I talk to about issues that make me happy or sad	
Original version	3. My family really tries to help me.	

Forward translation	My family try by all means to help me
Original version	4. I get the emotional help & support I need from my family.
Forward translation	I get help and support that I need from my family when I have something troubling my mind
Original version	5. I have a special person who is a real source of comfort to me.
Forward translation	There is a special person who consoles me in difficult times
Original version	6. My friends really try to help me.
Forward translation	My friends try by all means to assist me
Original version	7. I can count on my friends when things go wrong.
Forward translation	I depend on my friends when things go wrong
Original version	8. I can talk about my problems with my family.
Forward translation	I can discuss my problems with my family
Original version	9. I have friends with whom I can share my joys and sorrows.
Forward translation	I have friends with whom I share my joys and sorrows.
Original version	10. There is a special person in my life who cares about my feelings.
Forward translation	There is a very special person in my life who cares about how I feel
Original version	11. My family is willing to help me make decisions.
Forward translation	My family is ready to help me make choices
Original version	12. I can talk about my problems with my friends.
Forward translation	I discuss my problems with friends.

11.13.8 Cognitive debriefing

11.13.8.1 Sample demographic characteristics

As shown in

Table 11.4, most caregivers were female, educated, mothers, married, unemployed, with a mean age of 33.4 years (SD 6.5) and had offered care for a median time of 17 months.

Table 11.4: MSPSS Cognitive debriefing sample characteristics

Variable	Attribute	Frequency, n (%)
Gender	Female	15 (93.8)
	Male	1 (6.3)
Level of education	None	2 (12.5)
	Primary	3 (18.8)
	Secondary	7 (43.8)
	Tertiary	4 (25)
Place of residence	Urban	8 (50)
	Rural	8 (50)
Relationship to child	Mother	12 (75)
	Aunt	3 (18.8)
	Father	1 (6.3)
Marital status	Married	12 (75)
	Divorced	3 (18.8)
	Unknown	1 (6.3)
***Caregiver age	Mean (SD)	33.4 (SD 6.5) years
***Duration of caregiving	Median [$Q_1 - Q_3$]	17 [10-39] months
	Formally employed	4 (25)
	Housekeeper	2 (12.5)
	Informally employed	2 (12.5)
	Unemployed	8 (50)
Financial situation	Very inadequate	1 (6.3)
	Inadequate	5 (31.3)
	Neutral	8 (50)
	Adequate	2 (12.5)

N=16

11.13.8.2 Distribution of scores on the MSPSS

There was a strong, and significant correlation between scores on the English and Shona versions of the MSPSS, Rho=.996, p<.001. (See the scores distribution in Table 11.5.

Table 11.5: Distribution of scores on the English and Shona versions of the MSPSS

Question	Language	Strongly disagree (1)	Disagree (2)	Neutral (3)	Agree (4)	Strongly agree (5)
1. There is a special person who is around when I am in need	English		1	3	5	7
	Shona	1	3	1	6	5
2. There is a special person with whom I can share joys and sorrows	English	1	3	1	7	4
	Shona	1	1	4	5	5
3. My family really tries to help me.	English	1	1	4	5	5
	Shona	1	1	4	4	6
4. I get the emotional help & support I need from my family.	English	1	1	3	5	6
	Shona		2	3	5	6
5. I have a special person who is a real source of comfort to me.	English		1	4	5	6
	Shona	2	2	6	3	3
6. My friends really try to help me.	English	2	1	7	3	3
	Shona	1	5	4	4	2
7. I can count on my friends when things go wrong.	English	1	5	3	5	2
	Shona	1	3	1	8	3
8. I can talk about my problems with my family.	English	1	3	3	4	5
	Shona		4	3	7	2
9. I have friends with whom I can share my joys and sorrows.	English		3	4	8	1
	Shona	1	1	3	6	5
10. There is a special person in my life who cares about my feelings	English	1	1	3	5	6
	Shona	1	3	4	4	4
11. My family is willing to help me make decisions.	English	1	3	3	6	3
	Shona	1	2	5	5	3
12. I can talk about my problems with my friends	English	1	2	5	6	2
	Shona	1	2	5	6	2

N=16

11.13.8.3 Satisfaction with questionnaire

As outlined in Table 11.6 below, most respondents were satisfied with the instructions, clarity, response categories and relevancy of items on the MSPSS. However, three (3) respondents had challenges in understanding item 5 given the Shona variations of the term "decisions". We integrated alternatives suggested by caregivers in the harmonisation of the item.

Table 11.6: Respondents satisfaction with MSPSS questionnaire

Attribute	Strongly disagree (1)	Disagree (2)	Neutral (3)	Agree (4)	Strongly agree (5)
Instructions clarity				**9**	**7**
Questions clarity		**1**	1	6	8
Response categories			2	7	7
Questions relevancy to caregiving situation				7	9
Cultural relevancy		**1**	2	8	5

N=16

11.13.9 Discussion

We employed a robust translation process, and we believe this resulted in an accurate translation. Translators worked independently, were fluent in both source and target language and had varied experiences in translating PROs as we strived to gain a colloquial and conceptually equivalent translation [458,466,607-609]. Zimbabwe is a multilingual country with English being the official national language [587,588,610-615]. Shona and Ndebele are the predominant endoglossic languages as they are spoken by 80% and 16% of the population respectively, and there are also 14 official minority languages [587,588,610-612]. Due to linguistic diversity, urbanisation and modernisation, the Shona language has evolved, as such, the younger generation has been reported to perform poorly in indigenous languages [587,588,610-612]. The attainment of a simple and conceptually equivalent translation was thus crucial. For example, in reconciling forward translations of item 5, we dropped expressions which were deemed idiomatically complicated as they were potentially confusing to the younger generation of carers especially those dwelling in urban areas. Previous studies have shown that most Zimbabwean urban dwellers are bilingual and converse in "Shonglish" a lingo for blended English and Shona [610] thus were likely to have difficulties in understanding complex Shona expressions. Furthermore, the supremacy and dominance of English in both print and digital media and entertainment has somewhat stifled the development of indigenous languages [587-591,610-617]. For instance, Shona grammar in secondary school is taught in English [587,588,616], and it was important to take note of the intricacies and complexities of the Shona language during the translation

and adaptation process. Additionally, in some spheres, scholars and parents have expressed the sentiment that learning of indigenous languages such as Shona to be "…sheer waste of time" [587].

The involvement of the developer was also critical in the translation and adaptation process [3,53]. For instance, we could not find idiomatic equivalent expressions for the terms "feelings" and "emotion" and harmonisation of items 2, 4 and 10 was slightly problematic as translations were lengthy and contained some jargon. The difficulty in harmonisation can be attributed to the existence of diglossia in the Shona language. Diglossia can be defined "…as the differential and preferential use of a language or a dialect…" [589]. For example, among the 13 Shona dialects, there have been reports of preferential usage of the Zezuru and Chikaranga dialects in the teaching of the Shona language [587-591]. Additionally, we dropped ambiguous words for example, for item 1, we dropped the translation with the term "dambudziko" an equivalent to "problem" given its diverse conceptual meaning, i.e. it can mean "need", "trouble", "grief" or "challenge". Despite the slight idiomatic and grammatic discrepancies during the backward translation review, the developer confirmed the accuracy of the backward translation as it was conceptually equivalent to the original version.

Qualitative feedback from caregivers and a high correlation between scores on the English and Shona versions [Rho=.996, p<.001] further confirmed the accuracy of the translation during the cognitive debriefing phase. We also incorporated alternative expressions for the term "decision" on item 5 as some caregivers had expressed difficulty in understanding the term. The ambiguity can be attributed to diglossia and linguistic variation of the Shona language [587-591]. Nevertheless, this explains the need to involve target users in the translation and adaptation process to increase both face and content validity [3,53].

11.13.10 Conclusion

We applied a robust translation process, and we believe this yielded an accurate and conceptual equivalent MSPSS-Shona version. However, it is important to further evaluate other psychometric properties such as structural and known group-validity and reliability by administering the survey to a large group of caregivers. Further, there is a need to test the structural validity of the MSPSS-Shona version using item response theory techniques to increase the psychometric robustness of the MSPSS-Shona version further as recommended from our prior systematic review [53].

11.14 Appendix 14: English Version of the Multidimensional Scale of Perceived Social Support (MSPSS)

Instructions: We are interested in how you feel about the following statements.

Read each statement carefully. Indicate how you feel about each statement.

Circle the "1" if you Very Strongly Disagree

Circle the "2" if you Strongly Disagree

Circle the "3" if you Mildly Disagree

Circle the "4" if you are Neutral

Circle the "5" if you Mildly Agree

Circle the "6" if you Strongly Agree

Circle the "7" if you Very Strongly Agree

Item	Very Strongly Agree	Strongly Agree	Mildly Agree	Neutral	Mildly Disagree	Strongly Disagree	Very Strongly Disagree
1.There is a special person who is around when I am in need.	7	6	5	4	3	2	1
2. There is a special person with whom I can share my joys and sorrows.	7	6	5	4	3	2	1
3. My family really tries to help me	7	6	5	4	3	2	1
4. I get the emotional help and support I need from my family.	7	6	5	4	3	2	1
5. I have a special person who is a real source of comfort to me.	7	6	5	4	3	2	1
6. My friends really try to help me	7	6	5	4	3	2	1
7. I can count on my friends when things go wrong.	7	6	5	4	3	2	1
8. I can talk about my problems with my family.	7	6	5	4	3	2	1
9. I have friends with whom I can share my joys and sorrows.	7	6	5	4	3	2	1
10. There is a special person in my life that cares about my feelings	7	6	5	4	3	2	1
11. My family is willing to help me make decisions	7	6	5	4	3	2	1
12. I can talk about my problems with my friends	7	6	5	4	3	2	1

11.15 Appendix 15: Systematic review of translated versions of the MSPSS

Please click to open the full PDF file.

11.15.1 A systematic review of the psychometric properties of the cross-cultural translations and adaptations of the Multidimensional Perceived Social Support Scale (MSPSS)

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Health and Quality of Life Outcomes

REVIEW Open Access

A systematic review of the psychometric properties of the cross-cultural translations and adaptations of the Multidimensional Perceived Social Support Scale (MSPSS)

Jermaine M. Dambi[1,2*], Lieselotte Corten[1], Matthew Chiwaridzo[1,2], Helen Jack[3,4], Tecla Mlambo[2] and Jennifer Jelsma[1]

Abstract

Background: Social support (SS) has been identified as an essential buffer to stressful life events. Consequently, there has been a surge in the evaluation of SS as a wellbeing indicator. The Multidimensional Perceived Social Support Scale (MSPSS) has evolved as one of the most extensively translated and validated social support outcome measures. Due to linguistic and cultural differences, there is need to test the psychometrics of the adapted versions. However, there is a paucity of systematic evidence of the psychometrics of adapted and translated versions of the MSPSS across settings.

Objectives: To understand the psychometric properties of the MSPSS for non-English speaking populations by conducting a systematic review of studies that examine the psychometric properties of non-English versions of the MSPSS.

Methods: We searched Africa-Wide Information, CINAHL, Medline and PsycINFO, for articles published in English on the translation and or validation of the MSPSS. Methodological quality and quality of psychometric properties of the retrieved translations were assessed using the COSMIN checklist and a validated quality assessment criterion, respectively. The two assessments were combined to produce the best level of evidence per language/translation.

Results: Seventy articles evaluating the MSPSS in 22 languages were retrieved. Most translations [16/22] were not rigorously translated (only solitary backward-forward translations were performed, reconciliation was poorly described, or were not pretested). There was poor evidence for structural validity, as confirmatory factor analysis was performed in only nine studies. Internal consistency was reported in all studies. Most attained a Cronbach's alpha of at least 0.70 against a backdrop of fair methodological quality. There was poor evidence for construct validity.

Conclusion: There is limited evidence supporting the psychometric robustness of the translated versions of the MSPSS, and given the variability, the individual psychometrics of a translation must be considered prior to use. Responsiveness, measurement error and cut-off values should also be assessed to increase the clinical utility and psychometric robustness of the translated versions of the MSPSS.

Trial registration: PROSPERO - CRD42016052394.

Keywords: Multidimensional perceived social support, Translation, Adaptation, Validation, Reliability, Validity

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11.16 Appendix 16: Caregiver demographic details

Instruction: may you please indicate your responses by ticking in the appropriate box

Code ☐

1. What is your age? ☐ Years 2. What is your sex: Female ☐ Male ☐

3. Where do you stay? Rural area ☐ Urban ☐

4. How are you related to the child? Parent ☐

Grandparent ☐

Sibling ☐

If other, please specify……..

5. What is your marital status? Single ☐

Married ☐

Divorced ☐

If other, please specify……

6.What is your highest level of educational? Primary ☐

Secondary ☐

Tertiary ☐

If other, please specify……

7.What is your employment status? Unemployed ☐

Formally employed ☐

Informally employed ☐

Housekeeper ☐

Other, please specify……

11.17 Appendix 17: South African English version of the EQ-5D

By placing a tick in one box in each group below, please indicate which statements best describe your own state of health TODAY.

Mobility

I have no problems in walking about	❑
I have some problems in walking about	❑
I am confined to bed	❑

Self-Care

I have no problems with self-care	❑
I have some problems washing or dressing myself	❑
I am unable to wash or dress myself	❑

Usual Activities (*e.g. work, study, housework, family or leisure activities)*

I have no problems with performing my usual activities	❑
I have some problems with performing my usual activities	❑
I am unable to perform my usual activities	❑

Pain/Discomfort

I have no pain or discomfort	❑
I have moderate pain or discomfort	❑
I have extreme pain or discomfort	❑

Anxiety/Depression

I am not anxious or depressed	❑
I am moderately anxious or depressed	❑
I am extremely anxious or depressed	❑

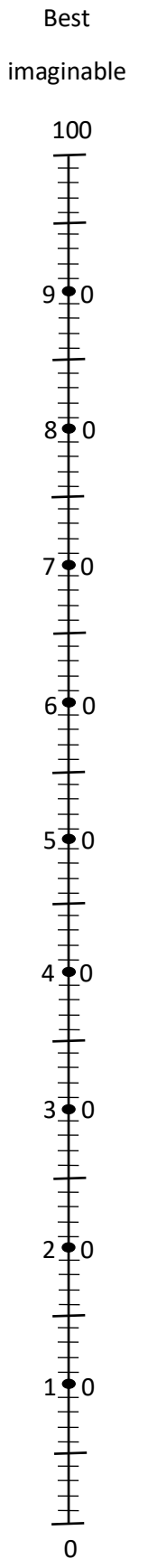
Compared with my general level of health over the past 12 months, my state of health today is:

Better	❑	PLEASE TICK
Much the same	❑	ONE
Worse	❑	BOX

To help people say how good or bad their state of health is, we have drawn a scale on which the best state you can imagine is marked 100 and the worst state you can imagine is marked 0.

We would like you to indicate on this scale, in your opinion, how good or bad your own health is today. Please do this by drawing a line from the box below to whichever point on the scale indicates how good or bad your state of health is today.

Your own state of health today



11.18 Appendix 18: EQ-5D – Shona version

Muchikwata chimwe nechimwe chemhinduro dzinotevera, isa mucherechedzo mukabhokisi kari kumucheto seizvi [✔] pamhinduro imwe chete yaunofunga kuti ndiyo inonyatsotsanangura utano hwako PARINHASI.

Kugona kufamba

Handinetseki kufamba ❑

Kufamba kunondinetsa ❑

Handigone kana nekufamba kose ❑

Kuzvishambidza

Ndinogona zvangu kuzvishambidza ❑

Handinyatsogona kuzvigezesa kana kuzvipfekedza ❑

Handigone kuzvigezesa kana kuzvipfedza ❑

Mabasa enguva dzose

Handinetseki nokuita mabasa angu andinowanzoita nguva dzose ❑

Ndinonetseka kuita mabasa angu andinowanzoita mazuva ose ❑

Handichagoni kuita mabasa angu andaiwanzoita mazuva ose ❑

Kurwadziwa/ Kusagadzikana

Handisi kurwadziwa ❑

Ndinorwadziwa zwangu zvishoma ❑

Ndinorwadziwa zvakanyanya. ❑

Kunetsekana mupfungwa / Kuremerwa

Hapana zvinondinetsa mupfungwa ❑

Ndine zvinodinetsa zwakati kuti ❑

Ndirikushushikana zvakanyanya ❑

Kana ndichienzanisa utano hwangu pamwedzi gumi nemiviri yapfuura neparinhasi,ndingati zvangu nhasi:

Zvava nani ❑ SARUDZA

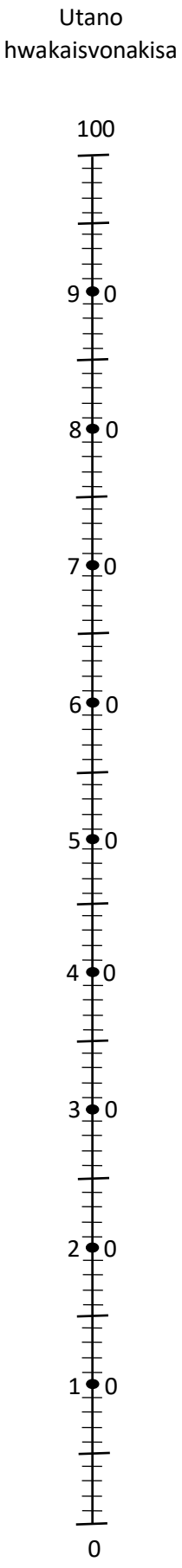
Zvakangofanana ❑ BHOKISI

Zvatonyanya ❑ RIMWE CHETE

Kuti tibatsire vanhu kuti vaone kunaka kana kushata kwakaita utano hwavo parinhasi, takupa chikero ichi chekupimisa nacho utano hwako. Chine nhamba dzinobvira pasi pana 0 kusvika kumusoro kuna 100. 0 anoratidza utano hwakadzikira hwemunhu anorwara zvakasvoipisa. 100 anoratidza utano hwakaisvonakisa hwemunhu asingarware.

Tinokumbira kuti unongedze nhamba pachikero apa yaunofunga kuti ndiyo inoratidza ipo chaipo pane utano hwako nhasi uno. Ita izvi nokunyora mutsetse unotangira kubva pachibhokisi chiri pazasi icho wakananga nechekurudyi uko kunechikero uchinoguma ipo chiapo pane nhamba yawasarudza yaunofungira kuti ndiyo chaiyo inoratidza pava neutano hwako nhasi.

Utano hwako nhasi uno.



11.19 Appendix 19: Caregiver Strain Index - English version

I am going to read a list of things that other people have found to be difficult. Would you tell me whether *any of these apply to you?*

	Yes = 1	No = 0
Sleep is disturbed (e.g., because . . . is in and out of bed or wanders around at night)		
It is inconvenient (e.g., because helping takes so much time)		
It is a physical strain (e.g., because of lifting in and out of a chair; effort or concentration is required)		
It is confining (e.g., helping restricts free time or cannot go visiting)		
There have been family adjustments (e.g., because helping has disrupted routine; there has been no privacy)		
There have been changes in personal plans (e.g., had to turn down a job; could not go on vacation)		
There have been emotional adjustments (e.g., because of severe arguments)		
Some behavior is upsetting (e.g., because of incontinence; . . . has trouble remembering things; or . . . accuses people of taking things)		
It is upsetting to find . . . has changed so much from his/her former self (e.g., he/she is a different person than he/she used to be)		
There have been work adjustments (e.g., because of having to take time off)		
It is a financial strain		
Feeling completely overwhelmed (e.g., because of worry about . . . ; concerns about how you will manage)		
Total Score		

11.20 Appendix 20: Caregiver Strain Index - Shona version

Ndichaverenga zvimwe zvezvinhu zvakaonekwa zvakaoma nevamwe vanhu. Mungadzidzewo here imi, kuti zvinokubatai Seiko.

	HONGU	**KWETE**
Kuvhiringidzwa kurara(semuenzaniso,nekuda kwekuti…anoti imwe nguva ari pamubheda kana anongo tenderera uri husiku).		
Zvinonetsa(semuenzaniso,sezvo uchisimudza murwere,panotoda samba rakawanda nekuzvipira zvakanyanya).		
Patova nekupesana kwemhuri(semuenzaniso unenge watosiya basa repamurungu,kutadza kana kenda kuzororo)		
Pave nekutsamwisana(semuenzaniso **zvakukonzera kupokana nekukakavadzana kusingapere)**		
Humwe hunhu hwake hunogumbura(semuenzaniso,anozviitira mubhurugwa)		
Zvinogumbura kuziva kuti…ashanduka zvikuru kubva zvaangaari,(sekuti,anenge ava munhu mutsva kubva zvaaiva ari)		
Zvinovhiringidza basa(sekuti unoramba ongotora mazuva ekusave pabasa akawanda)		
Ndinonzwa Kuremerwa kukuru(semuenzaniso,ndinonyanya kushushikana zvakanyanya uye ndinobatwa nekufunga neramangwana)		
Zvinokweva mari		
Ndinonzwa Kuremerwa kukuru		
TOTAL SCORE		

11.21 Appendix 21: The Shona Symptom Questionnaire

Musvondo rapfuura… **(During the course of the past week..)**	**Yes**	**No**
1)*..pane pamaimboona muchinyanya kufungisisa kana kufunga zvakawanda here?* ..did you have times in which you were thinking deep or thinking about many things?	1	0
2)*..pane pamaimbotadza kuisa pfungwa dzenyu pamwechete here?* ..did you find yourself at sometimes failing to concentrate?	1	0
3)*..maimboshatirwa kana kuita hasha zvenhando here?* ..did you lose your temper or getting annoyed over trivial matters?	1	0
4)*..maimborota hope dzinotyisa kana dzisina kunaka here?* ..did you have a nightmare or bad dreams?	1	0
5)*..maimboona kana kunzwa zvinhu zvangazvisingaonekwe kana kunzwikwa nevamwe here?* ..did you sometimes see or hear things which others would not see or hear?	1	0
6)*..mudumbu menyu maimborwadza here?* ..was your stomach aching?	1	0
7)*..maimbovhundutswa nezvinhu zvisina maturo here?* ..were you frightened by trivial things?	1	0
8)*..maimbotadza kurara kana kushaya hope here?* ..did you sometimes fail to sleep or lose sleep?	1	0
9)*..pane pamaimbonzwa muchiomerwa neupenyu zvekuti makambochema kana kuti makambonzwa kuda kuchema here?* ..were there moments when you felt life was so tough that you cried or wanted to cry?	1	0
10)*..maimbonzwa kuneta here?* ..did you feel run down (tired)?	1	0
11)*..pane pamaimboita pfungwa dzekuda kuzviuraya here?* ..did you, at times, feel like committing suicide?	1	0
12)*..mainzwa kusafara here mune zvamaiita zuva nezuva?* ..were you generally unhappy with things you were doing daily?	1	0
13)*..basa renyu raive rave kusarira mumashure here?* ..was your work lagging behind?	1	0
14)*..mainzwa zvichikuomerai here kuti muzive kuti moita zvipi?* ..did you feel you had problems in deciding what to do	1	0

11.22 Appendix 22: ZCCS- Final Version

ZIMBABWEAN CAREGIVER BURDEN SCALE

Instructions: Given below are some feelings/conditions that may be experienced by a person caring for a child with special needs/disability. Please select by **ticking** the most suitable response which best describes your experience. It is important that you answer **ALL** the questions. Please note that the term "child" refers to the child with special needs/ disability.

An example of how you can respond is as follows: I feel stressed by the demands of caring for the child	Yes, it's **very** true	Yes	Neutral	No ✔	No, **not at all**

#	QUESTION	RESPONSE				
The following questions relate to how **YOUR PHYSICAL HEALTH** may be affected by caregiving:						
1.	Considering my caregiving responsibilities, I do not have enough time for my basic needs such as having meals, bathing, etc.	Yes, it's very true	Yes	Neutral	No	No, not at all
2.	My sleep is disturbed (For example: the child I care for cries a lot and wakes me up at night)	Yes, it's very true	Yes	Neutral	No	No, not at all
3.	I feel tired and exhausted as a result of caring for the child.	Yes, it's very true	Yes	Neutral	No	No, not at all
4.	I usually feel body aches or discomfort in my body when caring for my child. (For example, I normally feel pain when lifting or carrying the child)	Yes, it's very true	Yes	Neutral	No	No, not at all
5.	In general, I feel that caregiving is a physical strain, i.e. it requires a lot of physical effort in performing the caregiving roles	Yes, it's very true	Yes	Neutral	No	No, not at all
The following questions relate to how providing care may affect YOUR FINANCES:						
6.	There have been adjustments in my income generation activities due to caregiving. (For example, I had to turn down a job, or I cannot engage in farming activities as much as I wish to)	Yes, it's very true	Yes	Neutral	No	No, not at all
7.	There has been an increase in our family expenses due to the child's condition.	Yes, it's very true	Yes	Neutral	No	No, not at all
8.	I worry that I am unable to provide special facilities and services needed by my child. (For example, providing for a special diet)	Yes, it's very true	Yes	Neutral	No	No, not at all
9.	In general, I feel that my child's condition places a financial strain on my family.	Yes, it's very true	Yes	Neutral	No	No, not at all
The following questions relate to how providing care may affect YOUR IMMEDIATE FAMILY:						
10.	Due to the responsibilities of caregiving, it is now difficult to plan for the future. (For example, it is difficult to plan to have other children)	Yes, it's very true	Yes	Neutral	No	No, not at all
11.	There has been a disruption or upset of relationships within the family. (For example, caring for my child often prevents me from attending to the needs of other family members)	Yes, it's very true	Yes	Neutral	No	No, not at all
12.	My spouse (significant other) does not help me with the care of this child. (For example, he/she does not support me financially in caring for my child)	Yes, it's very true	Yes	Neutral	No	No, not at all
13.	Parenting a child with a disability has caused more problems than I expected in my relationship with my spouse (significant other).	Yes, it's very true	Yes	Neutral	No	No, not at all
14.	Overall, I feel that my family has been negatively affected by my child's condition.	Yes, it's very true	Yes	Neutral	No	No, not at all
The following questions relate to how CONCERNS FOR YOUR CHILD may affect you:						
15.	I feel guilty about the potential cause(s) of my child's condition. (For example, something I did, or my partner did during pregnancy caused my child's condition)	Yes, it's very true	Yes	Neutral	No	No, not at all

16.	My child's sleeping and eating schedule were much harder to establish than I expected, and this worries me.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
17.	I worry about my child's future because of his/her condition. (For example, worrying about whether my child will be able to live independently as an adult)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
18.	My child seems to cry more often than most children and this upsets me.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
19.	I often feel embarrassed by my child's behaviour.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
20.	I worry that my child often gets insulted/ridiculed by other children.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
21.	I worry that other people don't know how to treat my child. (For example, I am even worried about bringing my child out to meet people)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
22.	I often face embarrassing situations when travelling with the child.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
23.	My spouse (significant other) helps me with the care of this child. (For example, he/she supports me financially in caring for my child)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
24.	I worry that my child is not able to communicate as I would have expected.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
25.	I feel sad that my child cannot do anything by him/herself like other children. (For example, playing, toileting, going to school etc.)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
26.	I worry about something bad happening to my child when s/he is out of my care.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
27.	My child falls ill from time to time, and this worries me.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
28.	I worry that my child's condition was caused by witchcraft or is a sign of bad luck/omen.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
29.	I worry that my child is not developing physically as other children of his/her age. (For example, he/she is underweight)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
The following questions relate to how providing care may affect YOUR PARTICIPATION IN THE COMMUNITY						
30.	Because of caregiving, it is difficult to keep in contact with relatives and friends. (For example, I rarely get the opportunity to visit my relatives)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
31.	It is very difficult to find suitable accommodation (For example, I must constantly move from one place to another as some landlords do not accept children with disabilities at their homes)	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
32.	I feel that I don't have enough time for your own interests/hobbies because of the amount of time I spend caregiving.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**
The following question relates to how YOU OVERALL view the caregiving process:						
33.	Overall, I feel completely overwhelmed by the caregiving role.	**Yes, it's very true**	**Yes**	**Neutral**	**No**	**No, not at all**

11.23 Appendix 23: Demographic questionnaires

11.23.1 Caregiver demographic questionnaire

Unique caregiver Code ☐

1. What is your age? Years 2. What is your sex: Fema ☐ Male ☐

3. Where do you stay? Urban area ☐ Rural area ☐

Relationship to child:

- Parent ☐
- Grandparent ☐
- Sibling ☐
- Employed caregiver ☐
- Other, please specify…….

What is your educational level?

- Primary ☐
- Secondary ☐
- Tertiary ☐
- If other, please specify……

What is your income level?

- Very inadequate ☐
- Inadequate ☐
- Neutral ☐
- Adequate ☐
- Very adequate ☐

11.23.2 Childs' demographic questionnaire

Age		GMFCS level	
Gender		Type of CP	

11.24 Appendix 24: Other publications arising from and related to the thesis

Click on publication to expand

11.24.1 Caring for a child with Cerebral Palsy: The experience of Zimbabwean mothers

Caring for a child with Cerebral Palsy: The experience of Zimbabwean mothers

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Background: The needs of caregivers of children with disability may not be recognized despite evidence to suggest that they experience increased strain because of their care-giving role. This strain may be exacerbated if they live in under-resourced areas.

Objectives: We set out to establish the well-being of caregivers of children with Cerebral Palsy (CP) living in high-density areas of Harare, Zimbabwe. In addition, we wished to identify factors that might be predictive of caregivers' well-being. Finally, we examined the psychometric properties of the Caregiver Strain Index (CSI) within the context of the study.

Method: Caregivers of 46 children with CP were assessed twice, at baseline, and after three months, for perceived burden of care and health-related quality of life (HRQoL) using CSI and EQ-5D respectively. The psychometric properties of the CSI were assessed post hoc.

Results: The caregivers reported considerable caregiver burden with half of the caregivers reporting CSI scores in the 'clinical distress' range. Many of the caregivers experienced some form of pain, depression and expressed that they were overwhelmed by the care-giving role. No variable was found to be associated with clinical distress. The CSI demonstrated good overall internal consistency (Cronbach's Alpha = 0.8), stability over time ($Z = 0.87$, $p = 0.381$) and was significantly and negatively correlated with the EQ-5D VAS (Spearman's rho = −0.33, $p = 0.027$), demonstrating concurrent validity.

Conclusion: Caregivers must be monitored routinely for their level of distress and there is an urgent need to provide them with support. The CSI is likely to be a valid measure of distress in this population.

Introduction

Problem statement

Cerebral Palsy (CP) is the most common physically disabling paediatric condition globally (Gagliardi *et al.* 2008). The prevalence in Zimbabwe is estimated to be at 1.55 per 1000 in rural areas and 3.3 per 1000 in urban areas (Finkenflugel *et al.* 1996). It is characterised by multiple impairments and functional limitations; essentially, it is a group of disorders that affects the development of movement and posture (Aisen *et al.* 2011). In Harare, the capital city of Zimbabwe, there are very few opportunities to place young children in appropriate day-care facilities, particularly those with severe disabilities from families with limited financial resources. Rehabilitation is provided on an intermittent basis by state institutions but caregivers, mostly mothers and sometimes members of the extended family do the day-to-day care.

Even in contexts in which there may be more supportive care available, caring for a child with physical disabilities may have a negative impact on the health and well-being of caregivers (Moster *et al.* 2010; Raina *et al.* 2005). CP is associated with multiple impairments and consequently, it is associated with multiple activity limitations and participation restrictions (Deepthi & Krishnamurthy 2011; Martin *et al.* 2005; Moster *et al.* 2010; Sharan *et al.* 2012).

Although the child with a disability may be a source of joy to the parents, their special needs may add to the emotional, physical and financial strain inherent in raising children, particularly in under-resourced settings. In addition, there is a consensus that long-term care-giving may affect physical, social and emotional health of the caregivers, their well-being, marital relationships, and employment and financial status (Davis *et al.* 2009; Yilmaz, Erkin & Ezke 2013). These added concerns may lead to subsequent deterioration of health and health-related quality of life of the caregivers (HRQoL) (Berker & Yalçin 2008; Deepthi & Krishnamurthy 2011; Jones *et al.* 2007; Martin *et al.* 2005; Oh & Lee 2009; O'Shea 2008; Rodrigues dos Santos *et al.* 2009). It is obviously also in the best interest of the child that strain in their caregivers be recognized and appropriate

11.24.2 A critical evaluation of the effectiveness of interventions for improving the well-being of caregivers of children with cerebral palsy: a systematic review protocol

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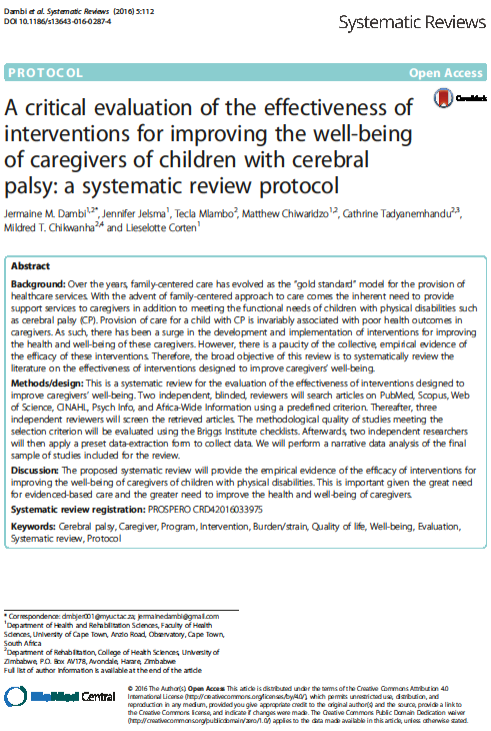

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Systematic Reviews

PROTOCOL Open Access

A critical evaluation of the effectiveness of interventions for improving the well-being of caregivers of children with cerebral palsy: a systematic review protocol

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Abstract

Background: Over the years, family-centered care has evolved as the "gold standard" model for the provision of healthcare services. With the advent of family-centered approach to care comes the inherent need to provide support services to caregivers in addition to meeting the functional needs of children with physical disabilities such as cerebral palsy (CP). Provision of care for a child with CP is invariably associated with poor health outcomes in caregivers. As such, there has been a surge in the development and implementation of interventions for improving the health and well-being of these caregivers. However, there is a paucity of the collective, empirical evidence of the efficacy of these interventions. Therefore, the broad objective of this review is to systematically review the literature on the effectiveness of interventions designed to improve caregivers' well-being.

Methods/design: This is a systematic review for the evaluation of the effectiveness of interventions designed to improve caregivers' well-being. Two independent, blinded, reviewers will search articles on PubMed, Scopus, Web of Science, CINAHL, Psych Info, and Africa-Wide Information using a predefined criterion. Thereafter, three independent reviewers will screen the retrieved articles. The methodological quality of studies meeting the selection criterion will be evaluated using the Briggs Institute checklists. Afterwards, two independent researchers will then apply a preset data-extraction form to collect data. We will perform a narrative data analysis of the final sample of studies included for the review.

Discussion: The proposed systematic review will provide the empirical evidence of the efficacy of interventions for improving the well-being of caregivers of children with physical disabilities. This is important given the great need for evidenced-based care and the greater need to improve the health and well-being of caregivers.

Systematic review registration: PROSPERO CRD42016033975

Keywords: Cerebral palsy, Caregiver, Program, Intervention, Burden/strain, Quality of life, Well-being, Evaluation, Systematic review, Protocol

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11.24.3 An evaluation of psychometric properties of caregiver burden outcome measures used in caregivers of children with cerebral palsy: a systematic review protocol

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Systematic Reviews

PROTOCOL Open Access

An evaluation of psychometric properties of caregiver burden outcome measures used in caregivers of children with cerebral palsy: a systematic review protocol

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Abstract

Background: Cerebral palsy (CP) is the most common, life-long paediatric disability. Taking care of a child with CP often results in caregiver burden/strain in the long run. As caregivers play an essential role in the rehabilitation of these children, it is therefore important to routinely screen for health outcomes in informal caregivers. Consequently, a plethora of caregiver burden outcome measures have been developed; however, there is a dearth of evidence of the most psychometrically sound tools. Therefore, the broad objective of this systematic review is to evaluate the psychometrical properties and clinical utility of tools used to measure caregiver burden in caregivers of children with CP.

Methods/design: This is a systematic review for the evaluation of the psychometric properties of caregiver burden outcome tools. Two independent and blinded reviewers will search articles on PubMed, Scopus, Web of Science, CINAHL, PsychINFO and Africa-Wide Google Scholar. Information will be analysed using predefined criteria. Thereafter, three independent reviewers will then screen the retrieved articles. The methodological quality of studies on the development and validation of the identified tools will be evaluated using the four point COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist. Finally, the psychometric properties of the tools which were developed and validated from methodological sound studies will then be analysed using predefined criteria.

Discussion: The proposed systematic review will give an extensive review of the psychometrical properties of tools used to measure caregiver burden in caregivers of children with CP. We hope to identify tools that can be used to accurately screen for caregiver burden both in clinical setting and for research purposes.

Systematic review registration: PROSPERO CRD42015028026.

Keywords: Cerebral Palsy, Caregiver burden/strain, Outcome measurement, Psychometric properties, Evaluation, Systematic review, Protocol

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11.24.4 Use of patient-reported outcomes in low-resource settings – lessons from the development and validation of the Zimbabwe Caregiver Burden Scale

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Use of patient-reported outcomes in low-resource settings — lessons from the development and validation of the Zimbabwean Caregiver Burden Scale

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